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A Systems Approach to Understanding Families of Children with Intellectual  
Disability and/or Autism.

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## Declarations

This thesis has not been submitted for an award or degree at any university or institution. Where material has been derived from other sources, full bibliographical information has been provided.

The work submitted is the result of my own investigations conducted under the supervision of Professor Richard Hastings and Dr Vasiliki Totsika. Additional supervisory support was also provided during the analysis and drafting stage of Chapter 4 by Dr Tom Bailey, a Research Fellow in CEDAR, Warwick.

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## Summary

While it is widely acknowledged that all family members have a role to play in raising a child with Intellectual Disability (ID), research in the field has focused on the wellbeing of mothers and mother-child relationships. Working within a Family Systems theoretical framework, this thesis has posed systemic questions and examined subsystems and members of the family unit, neglected in existing research, in order to develop a more holistic understanding of families of children with ID. It has also contributed large-scale survey data on families, something which is limited in the UK. In Chapter 1 a review of Family Systems Theory (FST) and existing systems-informed studies was undertaken, highlighting many unexplored avenues for research and outlining important methodological considerations. Four empirical studies then followed (Chapters 2,3,4,5). In the first study (Chapter 2) the relationship satisfaction of parents of children with Autism Spectrum Disorder (ASD) was explored. Chapter 3 then presented an investigation of the psychological wellbeing of fathers of children with and without ID. Chapters 4 and 5 presented findings based on the primary data collected for the Cerebra 1000 Families study, a large-scale survey of UK families of children with ID aged 4-15 years. Chapter 4 investigated whether mothers' perceptions of the functioning of three different family subsystems related to their overall rating of family functioning. The final study (chapter 5) then explored the psychological wellbeing of single mothers. In Chapter 6 the findings from the four empirical studies were discussed along with their implications for theory, practice and future research. Overall this thesis has contributed new knowledge in relation to the family systems of families of children with ID, and has rejected the prevailing narrative that raising a child with ID is always a negative experience.

## **Chapter 1: An Introduction to the Systems Approach to Family Disability and Background to the Thesis**

## **Context and Definitions**

It is estimated that around 1% of the global population have an Intellectual Disability (ID), with highest prevalence estimates observed in children and adolescents (Maulik, Mascarenhas, Mathers, Dua, & Saxena, 2011). ID is currently characterised by significant limitations in intellectual functioning and adaptive behaviour which emerges during the developmental period and has a lasting effect on development (Carulla et al., 2011). ID often also co-occurs with Autism Spectrum Disorders (ASD) (Tonnsen et al., 2016), currently defined (DSM-5) as a life-long developmental disability where individuals present difficulties with social communication and interaction, and have restrictive and repetitive behaviours and interests (Volkmar & McParland, 2014). Approximately 30% of children with ID in England are estimated to have autism (Emerson & Baines, 2010).

Having a disability is not just experienced by the individual but those who care for and about the person with the disability (Blacher, Neece, & Paczkowski, 2005). Despite this, research has not always considered the child within the context of their family or fully explored the impact that raising a child with ID can have on family units and members. This thesis aims to develop a better understanding of families of children with ID to inform theory and plug gaps in knowledge. There was also a desire from the outset for the findings to be useful for families and contribute to evidence-based support.

## **The Need for a Systems Approach in ID research**

Researchers now recognise the impact of raising a child with ID on all family members and embrace systematic ways of working. Prior to this, there had been a tendency for research to focus attention on mothers of children with disabilities (Seligman & Darling, 2007), with an overwhelming body of work focusing on maternal adjustment (Blacher & Hatton, 2007) and mother-child reciprocal effects (Hastings, Petalas, Jones, & Totsika, 2014). As such, mothers' experiences have been taken to be representative of the whole family (Cridland, Jones, Magee & Caputi, 2014). While this may reflect the likelihood that a mother is the child's

primary caregiver (Seligman & Darling, 2007), we know that in many cases “families are more than just mothers” (Blacher & Hatton, 2007, p.535) and researchers in the ID field have begun to include analyses with other family members such as fathers and siblings. It is important however to recognise that there are assumptions about what a family ‘is’ (i.e. mother, father and two children), and that in reality family composition is diverse and context-bound. In the thesis ‘family’ was defined as immediate family members living in the same household. The nature of the data collected meant that analyses did focus on mothers and fathers of children with ID/autism, the child with ID/autism, and a sibling, however reports from grandmothers were available and included in chapter 3, in absence of a mother response. In addition to family composition, it is important that we begin to understand the influence that each family member (including the child with ID) has on one other. This way of thinking relates to the Family Systems Theory (FST).

### **Family systems theory.**

Family Systems Theory (FST) understands the family as an organised system, proposing that as human beings we do not live in isolation but in family units whereby individuals in that family have an influence on, and are influenced by, one another (Cox & Paley, 1997). This systemic way of thinking it is not bound to families or to psychology, but originates from General Systems Theories (GST) (Bertalanffy, 1968) which has been adopted within many other disciplines (Smith-Acuña, 2010). GST considers a ‘system’ to be a collection of parts which are interrelated and interdependent, proposing that we cannot understand the individual parts of any system without acknowledging the broader unit within which it resides (Bertalanffy, 1968). If one part of a system is changed, it is considered to have implications for other parts of the system and the system as a whole. Systems theories ultimately seek to understand phenomena within context (Smith-Acuña, 2010) and are not concerned with exploring purely linear cause and effect relationships (Cox & Paley, 1997).

GST was adopted by the family therapy movement in the 1960s/70s (Smith-Acuña, 2010) and key principles have been applied to the family (Cox & Paley, 1997). Family system approaches vary in regards to their focus: macro-level



approaches explore the way in which systems such as families interact with other systems, such as communities; whereas micro-level approaches explore interactions within the family itself (Cridland et al., 2014; Seligman & Darling, 2007). While FST was developed in the context of the general population, its core concepts can be applied to families of children with ID (Lounds & Seltzer, 2007). This thesis takes a micro-level approach, exploring relationships within the family of those with a child with ID.

### **System as an organised whole.**

A system is considered to be more than an assembly of its parts, it works as a 'meaningful whole' (Smith-Acuña, 2010). In the context of family systems this means that the family is not just a group of individuals, but individuals which exist within an emotional unit (Seligman & Darling, 2007). A system, like the family, cannot be understood by summing its parts (individuals) (Smith-Acuña, 2010) because individual family members interact with one another within the unit and with other systems outside of the family (Cox & Paley, 1997). FST rejects the idea of linear causality in favour of circular feedback: interactions which are reciprocal, in that they influence one another (Cox & Paley, 1997). Understanding the family unit in this way has implications for the way in which we understand and study families of children with ID. Firstly, it tell us that a child with a disability will have an impact on all family members and subsystems within the family and that we should be exploring outcomes at these various levels. Secondly, it informs us that family members and their interactions cannot be understood in isolation but in relation to other members of the family unit and the subsystems within which they live, which will now be discussed.

### **Subsystems.**

Systems are also considered to be hierarchical in nature (Smith-Acuña, 2010). Families are considered to be comprised of subsystems (Cox & Paley, 1997; Minichin, 1985): semi-independent systems which operate within the larger family system (Cridland et al., 2014). While dependent on individual family composition,

the three most common family subsystems or ‘dyads’ include: ‘parental’ (parent(s) and child), ‘marital’ (husband and wife), and ‘sibling’ (child and child).

The functioning of each subsystem is considered to have implications for other subsystems in the family unit. For example, if there is conflict in the spousal subsystem, this is purported to reverberate throughout the family, affecting the relationship between parents and children, and between children (Seligman & Darling, 2007). In the context of parenting a child with ID, there is reason to suggest that conflict in the parental subsystem between the child with ID and parents may have implications for the functioning of marital and sibling subsystems (Seligman & Darling, 2007). This interdependence of family structure also requires us to think about the dual roles that adults play in families as partners and parents (Cox & Paley, 1997).

### **Boundaries.**

Family systems are considered to be separated by boundaries – hypothetical divides which separate one system from another which are governed by implicit rules (Minuchin, 1985) such as who, when, and how family members participate in family life (Carroll, Olson & Buckmiller, 2007). These boundaries can be ‘external’ between the family and other systems, and ‘internal’ between subsystems within the family unit (Cridland et al., 2014; Seligman & Darling, 2007). Boundary rules within the family make it clear to family members who is included and excluded from certain subsystems within the family unit (Cridland et al., 2014). Subsystem permeability is linked to overall family functioning: boundaries may be ‘open’ with loosely defined family roles, or they may be ‘closed’ with rigid and restrictive family roles (Cridland et al., 2014). Optimally functioning families are considered to maintain a balance between these two types of subsystem permeability (Seligman & Darling, 2007). However, this may be a challenge to achieve when raising a child with ID. Additional caregiving demands may mean that there are more ‘open’ systems which result in boundary ambiguity: confusion over roles and responsibilities (Cridland et al., 2014). Boundary ambiguity is thought to have consequences for the way a family functions and interacts (Carroll et al., 2007). An example of boundary ambiguity may be where other children in the family assume caregiving and household responsibilities. Extensive investment in a child’s care by

a parent is also considered to lead to identity ambiguity. This is where a parent finds it difficult to view themselves as anything other than their child's parent and/or carer (O'Brien, 2007). These differences in boundary maintenance in the family may explain the diverse ways in which families and individual family members respond to raising a child with ID.

### **Adaptability.**

Systems need to be able to maintain their stable state and patterns (homeostasis) (Minuchin, 1985) and ensure equilibrium (a balance of inputs and outputs) (White, Klein, & Martin, 2015). To do this it needs to successfully adapt to changes and challenges in the environment. The opposite of adaptability, rigidity, is considered to be related to an increase in conflict and tension in the system (White et al., 2015). This is no different for families: change is considered to impact and reverberate around the family system (Walker, 2012) and so they need to adapt in times of stress to self-stabilise and ensure that it can continue to preserve its functioning and fulfil its purpose (Cox & Paley, 1997). The birth of a child with a disability is likely to test a family's stability and equilibrium, thus requiring it to adapt in a variety of ways (Seligman & Darling, 2007). This has led to researchers to pursue aspects of family functioning such as resilience in order to understand the adaption of families caring for a child with ID (McConnell & Savage, 2015).

Adaptability is also considered to be positively associated with the degree of 'variety' in a family system. Variety is defined as "the extent to which the system has the resources to meet new environmental demands or adapt to changes" (White et al., 2015, p.150). Systems which have access to more resources (i.e. have greater variety) may be more able to make the adaptations required to overcome the change in the environment (White et al., 2015). This may be particularly relevant to families with children with ID who may be less able to access such resources.

### **Family lifecycle.**

A family lifecycle refers to a series of life phases which are associated with different transitions and tasks for individuals in the family unit (Walker, 2012). An

example of a life phase may be the transition of a couple to parenthood. This is an important concept to consider when studying family functioning as it may be related to changes across time and in response to life events and transition periods (Cridland et al., 2014). Considering the family in the context of their life phase allows us to understand the needs of individuals within a family at particular points in time (Walker, 2012).

The family lifecycle may be of particular relevance when exploring the outcomes of families raising a child a disability. For example, the experiences of parents of a young or recently diagnosed child may be distinctly different from those where their child is older and/or received their child's diagnosis some time ago. The birth of a child with a disability is still considered to be a non-normative event (Lounds & Seltzer, 2007) which may have implications for how parents respond to this (potentially) unexpected life transition. Family demographics and compositions also change over time, which will have an impact on family subsystems, boundaries, and the way in which roles are assigned (Seligman & Darling, 2007) – which all have the potential to influence individual outcomes. This concept highlights the importance of considering the life phase that a family is in, particularly when conducting cross-sectional research but also in between data collection points in longitudinal research.

### **Overall evaluation of FST.**

FST has many merits, with both researchers and practitioners applying its principles in their research and practice, however some have questioned the extent to which systems theory more broadly can be regarded as a theory (White et al., 2015). This critique links to a broader discussion about what constitutes a 'theory', as theories themselves are "open to varying interpretations and are difficult to describe" (Hammond, 2018, p.1). Some suggest that a theory can be considered to a global explanation of a phenomenon or picture of how data fit together (Hammond, 2018), which would support that systems theory is a theory as it provides an overall explanation of systems (White et al., 2015) or in the case of FST, families. Further to this, the work of Cridland and colleagues (2014) describes how there is no single systems theory about families; suggesting that FST is just one of many theories

about ‘family’. Within this thesis, family systems theory was used more as a framework for justifying and building statistical models and hypothesising relationships. FST was also used as an explanatory framework when interpreting the results.

### **Review of System-Informed research in the ID field**

Systems-informed approaches are growing in popularity and complexity in the ID field (Hastings, 2016), yet there is still a dearth of research which includes more than two members in the family system or considers family systems beyond the parent-child subsystem. Studies which have explored more than two family members have typically included mothers, fathers, and the child with ID.

A study by Hastings (2003b) was one of the first to consider the effects of other family members (a child with autism and their spouse) on the stress reported by mothers and fathers. While mothers’ stress was found to be associated with both the child’s behaviour problems and their partner’s mental health problems, fathers’ stress was not associated with either of these factors. While this study had a small sample size ( $N=18$ ), its triadic design showed that not only might maternal wellbeing be affected by members of the family in addition to the child with ID, but that family members (in this case fathers) may not respond in the same way to having a child with ID or be affected by the same variables as mothers.

Mother-father differences in families of children with ID have continued to be examined, with mothers typically reporting greater levels of stress (Dabrowska & Pisula 2010; Herring et al, 2006), depression, and anxiety (Jones, Totsika, Hastings & Petalas, 2013; Lee, 2009; Vilaseca, Ferrer & Olmos, 2014), compared to fathers. Mothers have also been found to report more positive perceptions of their child (Hastings et al., 2005a; Hastings et al., 2005b; Jones et al., 2013; Kayfitz, Gragg, & Robert Orr, 2010; Vilaseca et al., 2014). However, there have been mixed findings in relation to predictors of maternal and paternal wellbeing. For example, while some have found that child behaviour problems predicted maternal but not paternal stress

(Hastings et al., 2005b), others have found that mothers and fathers were similarly affected by their child's behaviour problems (Herring et al., 2006; Jones et al., 2013).

Researchers in the ID field have also begun to consider the impact of other children in the family on the outcomes of family members. While there has been a substantial body of work in the ID field which has endeavoured to explore the impact of having a brother or sister with ID/autism on siblings (Kovshoff, Cebula, Tsai, Hastings, 2017), less attention has been given to the impact that siblings may indeed have on other members of their family. A study by Hall, Burns and Reiss (2007) explored the maternal distress of 150 families raising a child with Fragile X syndrome. Interestingly, mothers' symptoms of anxiety and depression appeared to be equally influenced by the behaviour problems of child with FXS and that of an unaffected sibling. Sibling adjustment has also been suggested to affect the outcomes of other children. A longitudinal and systems study of 60 families (mother, child, and a sibling) of children with autism (Hastings et al., 2014) was the first to find that a sibling's earlier behaviour problems were associated with increased behaviour problems for the child with autism two and a half to three years later.

### **Subsystem analyses.**

Family systems approaches have also led to research which has examined the subsystems of families of children with ID. One subsystem which has received attention is the marital/partner subsystem. As well as research which has examined the relationship stability (i.e. the likelihood of divorce or separation) of parents of children with ID (Hatton, Emerson, Graham, Blacher, & Llewellyn, 2010; Lundeby & Tøssebro, 2008; Risdal & Singer, 2004) and autism (Baeza-Velasco, Michelon, Rattaz, Pernon, & Baghdadli, 2013; Freedman, Kalb, Zablotzky, & Stuart, 2012; Hartley et al., 2010), there have been studies which have looked at the quality of the marital relationships of parents of children with ID who remain together. These have typically explored factors associated with marital quality and adjustment (Benson & Kersh, 2011; Robinson & Neece, 2015; Stoneman & Gavidia-Payne, 2006; Weitlauf, Vehorn, Taylor, & Warren, 2014) and marital quality as a predictor of individual wellbeing outcomes for mothers and fathers (Kersh, Hedvat, Hauser-Cram & Warfield, 2006; Norlin & Broberg, 2013).

There has also been work which has examined the interaction between marital subsystems and other subsystems, such as the parenting subsystem, within families of children with ID/autism. These studies have helped us to further understand the interconnected nature of family subsystems. For instance, the closeness of the mother-child relationship has been reported to have a significant effect on the marital satisfaction of mothers of adolescents and adults with ASD (Hartley, Barker, Baker, Seltzer, Greenberg, 2012). Marital satisfaction and interactions have also been found to be predict the parenting experiences (Hartley, Barker, Seltzer, Greenberg & Floyd, 2011) and parenting stress of mothers and fathers of children, adolescents and adults with ASD (Hartley, Papp & Bolt, 2016a). A study by Harley et al. (2016a) which used diary studies to capture the positive and negative marital interactions and levels of parenting stress of 176 married couples, found that negative marital interactions ‘spilled over’ into parenting experiences: a day with a high number of negative marital interactions was associated with a higher level of parenting stress for both mothers and fathers of children with ASD.

There are also increasing efforts to explore the sibling subsystem in the ID field. While the majority of research exploring the link between family context and siblings has tended to focus on the adjustment and outcomes of siblings (Stoneman, 2001), there has been research which has examined the impact of a child with ID on the quality of the sibling relationship. While further work is still needed, these studies have found that sibling relationships are not necessarily negatively implicated when a child in the family has ID (Rossister & Sharpe, 2001; Walton & Ingersoll, 2015), with some reporting more positive sibling relationships in families of children with autism and Down syndrome compared to parents of typically developing children (Kaminsky & Dewey, 2001; Roper, Allred, Mandleco, Freeborn, & Dyches, 2014).

Researchers have also begun to consider whether sibling relationships in families of children with ID are related to the child with ID and other members of the family. Child behaviour problems have been found to be associated with poorer sibling relationships in families of children with ASD (Petalas et al., 2012). The work of Hastings and Petalas (2014) which explored the self-rated reports of siblings

of children with autism aged 7-17 years, found that higher levels of behaviour problems for the child with ASD predicted decreased warmth/closeness and increased conflict in the sibling relationship. Broad Autism Phenotype (BAP) has also been found to be negatively associated with the sibling relationship, however only where there was maternal depression and high impact on the family (Walton & Ingersoll, 2015), highlighting the importance of taking into account parental wellbeing and caregiving outcomes. Other studies have also reported associations between the quality of the sibling relationship and parental reports of burden: Roper et al. (2014) found that parents of children with disabilities who reported increased levels of caregiving burden also reported less positive sibling relationships (Roper et al., 2014). Some studies have also explored associations between the sibling subsystem and other family subsystems such as the marital subsystem. The work of Rivers and Stoneman (2003) employed FST to study the sibling relationships of 50 families of children with autism, finding that increased stress in the marital subsystem was associated with less satisfied sibling relationships.

#### **Family-level variables: Family Quality of Life (FQOL).**

Far fewer studies have considered the impact of raising a child with ID on the family as a whole (Cox & Paley, 1997). Research on Family Quality of Life (FQOL) has increasingly become an area of interest in the ID field. Turnbull (2000) defines FQOL as “conditions where the family’s needs are met, and family members enjoy their life together as a family and have a chance to do things which are important to them” (as cited in Park et al., 2003, p.368). This realm of research has expanded upon established work on individual quality of life (QoL) (Hoffman, Marquis, Poston, Summers, & Turnbull, 2006), to explore how various domains of family life are impacted where there is a child in the family with a disability, and what the general perceptions are of family members about family life (Brown, MacAdam-Crisp, Wang & Iarocci, 2006). While initial work in this area focused on the development and measurement of the FQOL construct (Hoffman et al., 2006; Park et al., 2003; Poston et al., 2003; Summers et al., 2005), this is now well established with more studies beginning to use FQOL measures in their work.



A comparative study by Brown et al. (2006) which studied caregivers' reports of FQOL in families of children with Down syndrome, autism and children without ID with similar household compositions, found that families without a child with ID showed statistically higher levels of FQOL. However, it should be noted that this study did have a small sample size ( $N=69$ ) and closer examination shows that families of children with ID did not rate lower in all of the dimensions of FQOL (5 out of the 9 dimensions measured) (Hastings, 2016).

More recent work has begun to explore predictors of FQOL. A study by Pozo, Sarriá, and Brioso (2014) with parents of children with ASD found that autism severity and social support was a significant predictor of both mothers' and fathers' reports of FQOL. The behaviour problems of the child with ASD also had a negative effect on FQOL and psychological wellbeing, however this was an indirectly through parental reports of Sense of Coherence (SoC), defined as the extent to which an individual views their life as comprehensible, manageable and meaningful. Another study by McStay, Trembath, and Dissanayake (2014) of parents of children with ASD also reported that child behaviour problems predicted mothers' and fathers' FQOL ratings. However, a later study by Gardiner and Iarocci (2015) with caregivers of children and adolescents with ASD found that adaptive functioning, specifically daily living skills, emerged as a significant predictor of FQOL, even when controlling for child behaviour problems, the severity of the child's disability, and family income.

A review of system-informed studies in the ID field highlights the many unexplored avenues of research at individual, subsystem and broader family-unit levels. There is a clear need to pose new questions which go beyond the mother-child dyad and embrace the complexity of family life. This thesis has taken the opportunity to build on areas which have received an insufficient amount of attention to contribute new knowledge to the field.

### **Statistical Considerations in Family Research**

The complex nature of family relationships can pose challenges for research. To be able to conduct analyses which examine family subsystems and include multiple family members, a large sample size is required - which may be one of the reasons why systematic research is scarce and studies in the field of ID tend to be small-scale. To try to combat such an issue, data for this thesis drew upon existing larger-scale datasets and involved the generation of primary data via involvement in the Cerebra 1000 families study: a large UK-wide survey of parental caregivers of children with ID aged 4-15 years, which generated over 1000 responses.

There were a number of key considerations when designing the 1000 Families Study survey. Firstly, survey measures were carefully selected to ensure that comparisons could be made with previous studies in the ID field and the general population. It has been difficult to ascertain the true impact on families raising a child with ID due to a lack of measure consistency in the field and the comparative use of control groups rather than normative data (Hastings, 2016). Survey measures for the 1000 Families Study were also chosen based on their psychometric properties and whether they had standardised ways of scoring. Measures with clinical cut-off points were favoured as they could indicate whether an individual was experiencing symptoms within a clinical range. A number of studies have found that the majority of mothers and fathers with ID are *not* scoring above the clinical cut-off on measures including the Hospital Anxiety and Depression Scale (HADS; Zigmond & Snaith, 1983) (Hastings et al., 2005a; MacDonald, Hastings, & Fitzsimons, 2010). Hence, cut-off scores provide much needed context.

Important considerations also need to be made when conducting analyses within families of children with ID. The limited research conducting within-family analyses is perhaps linked to issues with circular causality (Cox & Paley, 1997) and the challenges in working with nested data (Cridland et al., 2014). As one of the main tenets of FST is that family members are interdependent (i.e. that they are more similar than different to one another) this violates assumptions of General Linear Models (GLM) whereby there is independence of observations (Jones et al., 2013). This is of particular importance when considering previous work in the field which has examined mother-father differences. Mothers and fathers are more likely to report in similar ways due to living in the same household (Jones et al., 2013) which

has implications for the findings of previous studies which have not controlled for interdependence. There has been a move in the field towards using more sophisticated statistical techniques such as Multilevel Modeling (MLM) to combat the complexities of analysing hierarchical data (Enders & Tofinghi, 2007).

Aforementioned work (Jones et al., 2013; Hartley et al., 2011; Hartley et al., 2016a, Hartley et al., 2016b) has effectively used MLM and this statistical technique was adopted in one of the empirical chapters in this thesis which examined mother-father differences.

## **Structure of the Thesis**

The thesis is comprised of four empirical research studies and a general discussion chapter. Each of the four empirical chapters are a standalone piece of work which have been, or are in the process of being, submitted for publication. The empirical studies are not designed to lead on from one another, however they are altogether contributing knowledge about the parents of children with ID and/or autism as a whole. While the analysis in chapter 2 was the only chapter to focus specifically on the parents of children with autism, parents of children with autism and ID were also included in the analyses performed in chapters 4 and 5.

Chapters 2 and 3 draw upon the data of two separate secondary datasets, and chapters 4 and 5 draw upon primary data collected for the Cerebra 1,000 Families Study (discussed below). Chapters 2 and 4 explicitly ask systemic questions, while chapters 3 and 5 focus on neglected members of the family system. Each chapter adopts its own methodology dependent on the research questions being asked.

Chapter 2 describes the analysis of data from an existing dataset of families of children with ASD aged 4-17 years old (Petalas et al., 2012). The chapter had two aims: first to expand our understanding of parental relationship satisfaction in families of children with ASD by exploring whether mothers and fathers in the same family reported similar or different levels of relationship satisfaction. Second, to explore whether relationship satisfaction was associated with parental and child

characteristics. Multilevel Modeling (MLM) was used to ensure that the analysis accounted for the interdependence of mother-father data.

Chapter 3 describes the secondary analysis of father data from the third wave of the Millennium Cohort Study (MCS), a UK population-representative longitudinal cohort study. The aim of the chapter was to contribute to the lack of research conducted on the wellbeing of fathers of children with ID. 256 fathers of a child identified as having ID, and 10,187 fathers without a child with ID, were compared on measures of individual wellbeing and parenting. The study then explored whether the presence of a child with ID was still a significant predictor of paternal wellbeing when controlling for a number of variables identified as significant correlates of wellbeing. Because the MCS is a population-representative dataset, the Complex Samples Procedure was employed in SPSS to ensure that appropriate design, sampling and attribution weights were applied to the data.

Chapters 4 and 5 describe data collected as part of the first wave of the Cerebra 1000 Families Study, which will now be outlined.

A core part of this PhD involved collaboration with the UK charity Cerebra, ([www.cerebra.org.uk](http://www.cerebra.org.uk)) to conduct a large-scale UK study of parental caregivers of children with ID in the UK. While the 1000 Families study is longitudinal, the analyses presented in chapters 4 and 5 are based on Wave 1 of the study which ran from 2014-2017. The rationale for the 1000 Families study was to further understanding of what it is like to raise a child with ID in the UK and to shape future support for this population. It aimed to be the largest study of families of children aged 4-11 years (later extended to 15 years, 11 months) with ID in the UK, involving at least 1000 parental caregivers. To take part in the study respondents needed to be a caregiver of a child with ID and living in the UK. The child with ID could also have other conditions in addition to ID such as autism and genetic syndromes. Within the total sample: 50.5% of children were reported to have Autism Spectrum Disorder (ASD), 31.% Global Developmental Delay and 15.1% Down syndrome. 38.7% of parents reported that children had other genetic syndromes with the most common syndromes including Fragile X syndrome, Angleman Syndrome, Williams syndrome and conditions related to chromosome disorders.

The study was targeted at all parental caregivers but we were keen to recruit fathers and two participants per family: one from a primary caregiver (which would most likely be the mother of the child) and one from a secondary caregiver (who could be a father, grandparent, or any other adult with caregiving responsibilities living in the household).

Participating in the study involved the completion of an online or paper survey which asked questions about the caregiver's family and their experiences, and took on average 20 minutes to complete (see Appendices E, p.191 and F, p.240). Primary caregivers of the child were also asked at the beginning of their survey whether they would be willing to take part in an optional telephone interview about their child (Vineland-II Adaptive Behaviour Scales assessment). This meant that there were two separate surveys and accompanying information sheets for primary and secondary caregivers, however these were identical in content other than a question in the primary caregiver survey which asked respondents whether they would also be willing to take part in the telephone interview.

Project inception was in 2014 where the research team designed the survey and submitted an application to the National Health Service Regional Ethics Committee Service (NRES West Midlands, ref: 15/WM/0267) (see Appendix B p.155). Ethical approval was granted by the committee in September 2015 (see Appendix A p.150) and the team started to recruit to the study in November 2015. The study had a multi-point recruitment strategy: Cerebra as key stakeholders and funders of the study distributed information to a database of their members (see Appendix J, p. 270) and advertised the study via postcards (see Appendix K, p. 271), their website, and social media platforms (Facebook and Twitter). A project website was also created and interest in the study was also generated via the Cerebra Family Research Group's Facebook and Twitter pages (see Appendix L, p. 272). Other relevant organisations also shared information about the study on their own online platforms. Members of the research team attended relevant parent support groups, events, and conferences, and distributed surveys via contacts in special schools. Information about the study was also distributed via snowball sampling as primary caregivers who had taken part in the telephone interview agreed to promote the study via their own networks. Because of the multiple recruitment avenues it was not

possible to ascertain exactly where participants had heard about the study, however we did observe a significant increase in survey responses on the two occasions that Cerebra sent out information about the study to their database of members. Part way through the recruitment stage it was decided that we would extend the maximum child age range from 11 years, 11 months to 15 years, 11 months. This was partly to boost recruitment but also due to interest from parents of children of adolescent age. This involved an ethics amendment (see Appendix H, p. 262) which was approved by the committee in February 2017 (see Appendix G, p. 259). The study closed in August 2017 once we had received over 1000 survey responses.

Chapter 4 describes an exploratory analysis which investigated whether three family subsystems (marital, parental, and sibling) were related to the perceptions of family functioning of mothers of children with ID, as no other studies have conducted such an analysis in the ID field. Structural Equation Models (SEM) were fitted to address this research question.

Chapter 5 explored the psychological wellbeing of single mothers of children with ID, as the majority of work in the ID field has focused on mothers in two-parent households. Regression analyses examined whether a range of mother, child, and family-level factors were related to maternal psychological distress (depression, anxiety) and life satisfaction.

The sixth and final chapter provides a general discussion summarising the findings from the four empirical studies. Implications are also discussed in relation to theory, practice, and future research.

**Chapter 2<sup>1</sup>: Parental Relationship Satisfaction in Families of Children with  
Autism Spectrum Disorder (ASD): A Multilevel Analysis**

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<sup>1</sup> A version of this chapter has been published as Langley, E., Totsika, V., & Hastings, R.P. (2017). Parental Relationship Satisfaction in Families of Children with Autism Spectrum Disorder (ASD): A Multilevel Analysis. *Autism Research*, 10, 1259-1268. doi: 10.1002/aur.1773

### **Abstract**

Caring for a child with Autism Spectrum Disorder (ASD) has been linked to a range of negative outcomes for parents but less is known about the putative impact upon the parental couple relationship. We investigated the relationship satisfaction of parents of children with ASD using multilevel modeling. Mothers and fathers (146 couples) reported on their relationship satisfaction, their own wellbeing, and the behaviour problems of the child with ASD and a sibling. Results indicated that mothers and fathers reported similar levels of relationship satisfaction and it was significantly and negatively associated with parental depression and the behaviour problems of the child with ASD. Relationship satisfaction was unrelated to the behaviour problems of a sibling, the number of children in the household, and family socioeconomic position (SEP). Further longitudinal research that captures a broader range of variables is required to build a theoretical understanding of relationship satisfaction in families of children with ASD. Current evidence suggests that early intervention routes targeting either child behaviour problems, parental mental health, or the couple relationship have the potential to benefit interconnected subsystems within the broader family system.

**Key words:** Couples, ASD, relationship satisfaction, multilevel modeling



## **Introduction**

Parents of a child with a disability may encounter challenges above and beyond those associated with parenting a typically developing child which may affect their relationship with their partner (Saini et al., 2015). Caring for a child with Autism Spectrum Disorder (ASD) has been linked to a range of negative outcomes for parents such as elevated levels of stress (Eisenhower, Baker, & Blacher, 2005), depression (Singer, 2006) and burden (Stuart & McGrew, 2009), however, less is known about the couple relationship in families of children with ASD (Saini et al., 2015). The additional practical, emotional and financial demands of raising a child with ASD may change the everyday life and long-term outlook of couples. They may have less time to spend with one another, struggle to cope with their own and their partner's response to having a child with a disability, and face challenges balancing their role as a partner and a parent (Brobst, Clopton, & Hendrick, 2009).

There is growing recognition from disability family researchers that systems thinking is required to develop a fuller understanding of the impact of children with ID on family members (Cridland et al., 2014; Hastings, 2016). Family Systems Theory (FST) is a theoretical framework which emphasises the systematic, interrelatedness and complexity of family relationships, supporting the view that one family member's experience, such as having a disability, has the potential to affect all others in the family system and all family subsystems (Cox & Paley, 1997; Seligman & Darling, 2007; Smith-Acuña, 2010). The quality of the couple relationship has been found to be a predictor of parental wellbeing (Hartley et al., 2016a; Kersh, Hedvat, Hauser-Cram, & Warfield, 2006; Norlin & Broberg, 2013), coping (Siman-Tov & Kaniel, 2011) and burden (Hartley et al., 2011) in families of individuals with ASD and Developmental Disabilities (DD).

### **Parental Relationship Stability**

Some researchers have studied the relationship stability (how likely parents are to divorce or separate) of parents of children with ASD (Freedman et al. 2012). Saini et al. (2015) identified 11 studies, two of which directly addressed divorce risk for parents of children with ASD. The first reported that the rate of divorce for

parents of children with ASD (23.5%) in the United States was almost twice that of the comparison group (13.8%) (Hartley et al., 2010). The second study explored the occurrence and timing of separation of parents of children with ASD and found that after 10 years, 89 out of 119 (74.8%) couples had remained together, suggesting a separation rate of 25.2% (Baeza-Velasco et al., 2013). Clearly more studies are required to further clarify the divorce rates among families of children with ASD, however studying relationship quality among couples allows us to better understand the experiences of parents who remain together and provide better intervention.

### **Parental Relationship Quality**

Research exploring relationship quality is complicated by the large number of terms used to describe the couple relationship such as relationship (or marital) satisfaction, quality, success, happiness, and dyadic adjustment which are often used interchangeably and captured by a variety of psychometric measures (Fincham & Rogge, 2010). Parents of children with ASD have been found to report lower levels of marital satisfaction compared to parents of children without disabilities (Brobst et al., 2009; Fisman, Wolf, & Noh, 1989; Gau et al., 2012; Lee, 2009; Santamaria, Cuzzocrea, Gugliandolo, & Larcan, 2012; Sim, Cordier, Vaz, & Falkmer, 2016), and these lower levels of marital satisfaction persist over time (McGrew & Keyes, 2014). Parents of children with ASD have also been found to report lower relationship satisfaction than parents of children with Intellectual Disability (ID) (Kwok, Leung, & Wong, 2014), and Down syndrome (Santamaria et al., 2012).

### **Gender Differences in Parental Relationship Satisfaction**

It is less clear whether mothers and fathers within the same family report similar or different levels of relationship satisfaction. We might expect in relation to FST that one parent is strongly influenced by the other and thus may report in the same way about their relationship – often referred to as ‘crossover’ (Gerstein, Crnic, Blacher, & Baker, 2009). Evidence in relation to ‘co-parenting’ a child with ASD also suggests that the effect on the individual parent may well be shared (Hock, Timm, & Ramisch, 2012). However, there are also reasons to believe that mothers and fathers may report differently about their relationship satisfaction because of

reported gender differences in psychological wellbeing (Jones et al., 2013; Lee, 2009). Gender differences could also reflect the direct impact that raising a child with ASD has on mothers as they are more likely to be the primary caregiver (Hartley, Mihaila, Otolara-Fadner & Bussanich, 2014) and may have to give up their working role for a caregiving one (Gray, 2003; Twenge, Campbell, & Foster, 2003). Qualitative evidence also suggests that work and time demands can affect fathers' ability to remain an engaged partner (Meadan, Stoner, & Angell, 2015). A few studies have explored the within-gender differences in relationship satisfaction of parents of children with ASD using the Dyadic Adjustment Scale (DAS). No significant gender differences have been reported on total DAS scores (Lee, 2009), however some differences have been reported on the degree of consensus (Lee, 2009) and dyadic satisfaction and affection expression (Gau et al., 2012) subscales of the measure.

### **Correlates of Parental Relationship Satisfaction**

There is evidence to suggest that individual-level factors such as parental mental health may be associated with relationship satisfaction. Parents of children with ASD typically experience elevated levels of stress (Brobst et al., 2009; Shtayermman, 2013) and depression compared to other groups (Abbeduto et al., 2004; Blacher & McIntyre, 2006; Gau et al., 2012; Lai, Goh, Oei, & Sung, 2015). A meta-analysis of depression levels for parents of children with DD by Singer (2006) reported higher effect sizes for parents of children with ASD compared to other conditions. Parents of children with ASD with increased levels of psychological wellbeing have been reported to experience greater marital quality (Benson and Kersh, 2011), whereas lower marital satisfaction has been associated with higher levels of depression (Benson & Kersh, 2011; Shtayermman, 2013; Weitlauf et al., 2014). A study by Timmons, Willis, Pruitt and Ekas (2016) which explored the daily relationship quality of mothers of children with ASD reported that for every unit increase in depressive symptoms, mothers were 1.03 times more likely to engage in conflict with their partner on a given day. It appears that an individual parent's level of wellbeing could be a significant correlate of parental relationship satisfaction.

In addition, child characteristics are related to relationship satisfaction. Child behaviour problems have been consistently associated with elevated levels of parental psychological distress (Bromley, Hare, Davison, & Emerson, 2004; Estes et al., 2009; Jones et al., 2013); parenting stress (Lecavalier, 2006); and depression (Abbeduto et al., 2004; Benson & Kersh, 2011; Orsmond, Lin, & Seltzer, 2007) for parents of children with ASD and reported to lower relationship satisfaction (Hartley et al., 2012). The needs of a child with behaviour problems are likely to place a constant demand on parents and challenge boundaries in the family unit (Tsibidaki, 2013). Maintaining boundaries between particular subsystems (such as couple and parental) is considered important in order to preserve their function, which may be more of a challenge where the behavioural needs of a child take priority (Tsibidaki, 2013).

A study conducted by Robinson and Neece (2015) of parents of children with DD (where 88% had a diagnosis of ASD) showed that parents with the lowest marital satisfaction had children who displayed the most behaviour problems. Correspondingly, Sikora et al. (2013) reported significant associations between externalising behaviours and marriage impact, and Benson and Kersh (2011) reported a negative association between marital quality and child problem behaviours, and a positive association between marital quality and child pro-social behaviours, in parents of children with ASD. There have been some studies that have failed to find an association between child behaviour problems and relationship satisfaction (Weitlauf et al., 2014), and the intensity of behaviour problems and relationship satisfaction (Brobst et al., 2009), however overall there is a strong case for exploring the relationship between child behaviour problems and parental relationship satisfaction.

When considering the impact of the child with ASD on parental relationship satisfaction it is prudent to also explore any potential impact of sibling behaviour, as they too are within the family system. Studies which have explored the impact of having a brother or sister with ASD on sibling outcomes have shown elevated levels of behavioural and emotional problems when compared to comparable normative groups (Hastings, 2003a; Verte, Roeyers, & Buysse, 2003). It is therefore imperative

to explore whether a sibling's behaviour impacts upon their parent's relationship satisfaction.

At present, there is no specific theoretical framework to identify factors that may be associated with parental relationship satisfaction in families of children with ASD. However there are models in general developmental psychology such as the Family Stress Model (FSM) which does explore relationship satisfaction in the general population, hypothesising that financial pressures raise individual symptoms of depression which can result in poorer couple relationships (Conger, Conger, & Martin, 2010). There have been some studies which have investigated income, education and employment or socioeconomic status (SES) more broadly and their longitudinal associations with the wellbeing of parents of children with disabilities. Benson and Kersh (2011) found that marital quality was significantly associated with family SES with mothers of children with ASD. Hartley et al. (2012) examined the marital satisfaction of mothers of adolescents and adults with ASD over a 7-year period and found that it was positively related to household income. In addition, the number of children in the family may also be an additional strain on parents' cognitive and financial resources. A study by Harper, Dyches, Haper, Roper, & South (2013) found a negative association between the number of children and relationship quality for both mothers and fathers of children with ASD.

### **Statistical Framework**

In addition to a lack of a guiding theoretical framework, there are conceptual analytical problems with much existing research on parental relationship satisfaction in families of children with ASD. Parent data are nested within couples within families and as such require more sophisticated statistical techniques. Multilevel Models (MLM) can account for interdependence within their analyses (Enders & Tofighi, 2007) and are considered to be more accurate in estimating error terms for individuals and groups (Krull & MacKinnon, 2001). A selection of studies in this area have used MLM to explore predictors of relationship quality for mothers (Timmons et al., 2016), parental satisfaction (Ekas, Timmons, Pruitt, Ghilain, Alessandri, 2015), and 'spillover' between marital interactions and parenting stress (Hartley et al., 2016a), in families of children with ASD. Given the limited amount

of research in this area which has utilised MLM and the importance of accounting for the nested structure of the data, an MLM analysis approach was adopted.

The aim of the present study was to explore parental relationship satisfaction in families where one child has ASD. The research asked the following key research questions:

- Do mothers and fathers of children with ASD report different levels of relationship satisfaction?
- Is parent relationship satisfaction associated with parental depression, and the behavioural and emotional problems of the child with ASD and/or a sibling? And are any such associations still present after controlling for other family factors (socioeconomic position, number of children in the family)?

## Method

### Participants

One hundred and forty-six mother-father couples participated in the research. The majority of the couples in the sample were biological parents of their child with ASD, with the remaining including one adoptive parent couple, one foster parent couple, and six couples that included a biological mother and a stepfather. Mothers were on average 42 years-old ( $SD = 4.88$ , Range = 26-53 years) and fathers 44 years ( $SD = 5.20$ , Range = 30-64 years). Seventy-three (54.9%) mothers and 60 (45.1%) fathers were educated to university degree level or higher, with 88 (42.1%) mothers and 121 (57.9%) fathers in employment at the time of the research. Modal household income in the sample was £25,000-35,000 (British pounds sterling; approximately \$30,000-\$45,000 US dollars). The majority of parents described their ethnicity as White British (Mothers = 95.8%, Fathers = 94.6%). Most families had two children living in the family home (61.9%).

A family socioeconomic position (SEP) variable was created where families were categorised into one of four groups depending on whether at least one parent was educated to degree level (scoring one), whether at least one parent was employed (scoring one), and whether the household income was above the mode of the sample of £25,000-£35,000 (if so, scoring one). Seven (4.8%) families were categorised into the lowest scoring group of zero (neither parent educated to university level, neither parent in employment, and a household income of less than £35,000). Thirty-eight families (26%) comprised the second lowest group, scoring one indicator of high SEP; thirty-six (24.7%) families had two high SEP indicators; and sixty-five families (44.5%), had three high SEP indicators.

Parents reported that 89 (61%) children with ASD had a diagnosis of autism and 57 (39%) were reported as having a diagnosis of Asperger's syndrome. All children in this study had received their diagnoses before the application of the fifth edition of The Diagnostic and Statistical Manual of Mental Disorders (DSM-V) criteria. One hundred and twenty-two (83.6%) children with ASD were male and 24 (16.4%) were female. The child with ASD was on average 10.56 years of age ( $SD =$

2.81, Range = 4-17 years) and the majority of children had received their ASD diagnosis at on average 3.67 years (Range = 0.33 months - 12.83 years). Parents were asked to provide information about any sibling between the ages of 4 and 17 years of age also living in the family who did not have a disability. If there was more than one sibling in this age range, the sibling closest in age to the child with ASD was selected. Seventy three (50%) of the identified siblings were male, and seventy-three (50%) were female. Siblings had a mean age of 10.49 (SD = 3.44, Range = 4-17 years). Seventy-three (50%) siblings were younger than the child with ASD, sixty-five (44.5%) were older, and eight (5.5%) were twins. One hundred and fifty-four (52.7%) siblings were the same gender as the child with ASD.

### **Parental Measures**

The Dyadic Adjustment Scale-7 (DAS-7) (Hunsley, Pinsent, Lefebvre, Jamestaner, & Vito, 1995) was used to measure the relationship satisfaction of couples (see Appendix M, p. 273). This short-form version of the full (32 item) DAS (Spanier, 1976) contains seven items. Respondents are asked to rate 6 items on aspects such as “philosophy of life”, “amount of time spent together” and “working together on a project” on a Five-point scale (0=*Always disagree* to 5=*Always Agree*) and then complete one global dyadic satisfaction item ranging from 1 to 7. The DAS-7 has been found to be as psychometrically reliable as the full version of the DAS in assessing marital adjustment in clinical and community samples (Hunsley et al, 1995), with a reported average internal consistency of .80 (Hunsley, Best, Lefebvre, & Vito, 2001). The DAS-7 has been successfully used to assess marital quality in research studies with parents of children with ASD (Benson & Kersh, 2001) and DD (Kersh et al., 2006). A total DAS-7 score used in the present study is generated by summing all items (1-37), with higher scores indicating higher relationship satisfaction. Scores can be used to categorise relationships into distressed (1-21) or adjusted (22-37). Internal consistency (Cronbach’s  $\alpha$ ) in the present study was .88 for mothers and .83 for fathers.

The depression scale of the Hospital Anxiety and Depression Scale (HADS; Zigmond and Snaith, 1983) was used to assess parents’ depression levels (see Appendix N, p. 274). The HADS has been used extensively as a rapid measure of



depression in clinical and general populations (Bjelland, Dahl, Haug, & Neckelmann, 2002). It has previously been used effectively with parents of children with ASD (Hastings, 2003b). Depression has been justifiably entered into the model as it has previously been a valid measure of wellbeing in studies of parents of children with ASD. However this is not the case for anxiety, which was therefore not included in the analysis. The HADS contains seven items assessing depression (HADS-D). Respondents rate items such as “I feel as if I’m slowed down” and “I look forward with enjoyment to things” on a Four-point scale: *Most of the time, a lot of the time, from time to time, or not at all*. The total depression score ranges from 0-21 with higher scores indicating more depressive symptoms. Scores can also be used to classify depression symptoms into normal (0-10) and abnormal (11-21). Internal consistency was very good (Cronbach’s  $\alpha$  depression: Mothers .80 and fathers .78).

### **Child Behavioural and Emotional Problems**

The Strengths and Difficulties Questionnaire (SDQ; Goodman, 1997) was completed by primary caregivers as a measure of the behavioural and emotional problems of their children (see Appendix R, p. 278). The SDQ was completed for the child with ASD, and for the sibling that was closest in age to the child with ASD. The 25-item scale generates scores for four problem domains: Emotional symptoms, conduct problems, hyperactivity, and peer problems; and a pro-social behaviour domain. Caregivers indicate how likely each statement applies to the target child on a Three-point scale: *Not true, somewhat true, very true*, based on their child’s behaviour over the past six months. Sample items from each subscale include: “Often unhappy, downhearted or tearful” (emotional symptoms); “Often has temper tantrums or hot tempers” (Conduct problems); “Constantly fidgeting or squirming” (hyperactivity); “Has at least one good friend” (peer problems); “Considerate of other people’s feelings” (prosocial behaviour). The SDQ is a valid brief measure that has been reported to effectively assess the psychological adjustment of children and adolescent in normative samples (Goodman, 2001) and in research with children with ASD (Totsika et al., 2011a, Totsika et al., 2011b) and the adjustment of their siblings (Hastings, 2003a). In the present study, the total problem difficulties score was used. A higher score is indicative of greater behavioural and emotional

difficulties. The scores can also be used to categorise problem behaviour levels into normal (0-13), borderline (14-16) and abnormal (17-40). Internal consistency (Cronbach's  $\alpha$ ) obtained for this total difficulties score for the child with ASD was .71 and for the sibling .77.

## **Procedure**

The data used for the present analysis were part of a larger ASD family research study (Petalas et al., 2012). Following ethical approval, invitations to families were given to a national ASD charity to distribute to their members. The number of families contacted at this stage is unknown since national and local groups distributed study information through direct contact, advertisements, and mailing lists. A total of 305 families who expressed an interest met the criteria (a primary caregiver present in the home, with a child with ASD between the ages of 4-17 years old). Both parents were mailed a postal survey and asked to complete it separately. 215 families returned completed questionnaire packs. The present research focused on the data from 146 mother-father couples from families of children with ASD and also at least one sibling.

### **Statistical analysis approach.**

Multilevel models were fitted to examine the association of relationship satisfaction and the parent, child and family variables. Data in the study were structured in a 2-tier hierarchical data structure with individual variation at Level 1 and family variation at Level 2. Level 1 variables included parent gender and parent scores on dyadic adjustment and depression levels. Level 2 variables included family measures (family SEP, number of children in the family, behaviour problems of the child with ASD, and behaviour problems of the sibling). In the current study the family unit was modelled as a random factor, all other variables were modelled as fixed.

A variance components (VC) covariance structure was used to estimate the model parameters. VC provides separate variance estimates for each random effect and is often used as the default covariance structure when there is only one random

effect (Heck, Thomas, & Tabata, 2014). Predictor and control variables in the study were grand-mean centered, with the exception of family SEP which was median-centered. Centering allows variables to be transformed into meaningful deviations around a fixed value (Heck et al., 2014) and can improve interpretation and accuracy when estimating parameters (Enders & Tofighi, 2007). Statistical analyses in the study were conducted using SPSS Statistics 22.0®.

## Results

### Descriptive Statistics

Table 2.1 presents the mean relationship satisfaction score for mothers and fathers and the proportion of scores falling into distressed and adjusted categories. The total relationship satisfaction mean was 21.80 with 55.4% of relationships classified within the ‘adjusted’ range.

*Table 2.1 Mean dyadic adjustment score and proportion of sample scoring in adjusted and distressed categories (mothers and fathers)*

Measure	Mean (SD)	Reported range	Distressed (0-21)	Adjusted (22-37)
DAS-7 (Total)	21.80 (6.05)	1-36	44.6%	55.4%
DAS-7 (Mothers)	21.52 (6.52)	1-36	44.6%	55.4%
DAS-7 (Fathers)	22.11 (5.46)	4-35	44.5%	55.5%

The mean for the seven depression items of the HADS was 6.84 with the majority of scores falling into the normal category (80.5%) (Table 2.2). Mothers were more likely to score above the clinical cut-off than fathers (Table 2.2) and there was a significant difference in the scores for mothers ( $M=7.54$ ,  $SD = 4.28$ ) and fathers ( $M= 5.96$ ,  $SD= 3.83$ );  $t(129)= 3.10$ ,  $p = .002$ , with mothers reporting higher HADS-D scores than fathers (Table 2.3). Table 2.4 shows the mean SDQ total problem score to be 21.26 for the child with ASD, with the majority categorised in the abnormal range (81%), and 9.22 for siblings, with the majority of scores in the normal category (77.9%).

*Table 2.2 Mean depression score and proportion of sample scoring in the normal and abnormal categories (mothers and fathers)*

Measure	Mean (SD)	Reported range	Normal (0-10)	Abnormal (11+)
HADS-D (Total)	6.84 (4.16)	0-20	80.5%	19.5%
HADS-D (Mothers)	7.54 (4.28)	0-17	41.3%	14.8%
HADS-D (Fathers)	5.96 (3.83)	0-20	39.3%	4.7%

*Table 2.3 Paired sample t-test of maternal and paternal depression scores*

Paired samples Test									
		Paired Differences							
					95% Confidence Interval of the Difference				
					Lower	Upper			
		Mean	Std. Deviation	Std. Error Mean			t	df	Sig. (2-tailed)
Pair 1	HADS-D Mothers -HADS-D Fathers	1.25262	4.59993	.40344	.45440	2.05083	3.105	129	.002

*Table 2.4 Mean strengths and difficulties score and proportion of sample scoring in SDQ categories (individually for child and a sibling)*

Measure	Mean (SD)	Reported range	Normal (0-13)	Borderline (14-16)	Abnormal (17-40)
SDQ Child	21.26 (6.46)	6-37	7.7%	11.3%	81%
SDQ Sibling	9.22 (6.86)	0-32	77.9%	7.0%	15.1%

### **Intercept-only Model**

In the first stage of the analysis an intercept-only model with no predictors (null model) was fitted to explore the estimated variance in relationship satisfaction scores within (Level 1) and between families (Level 2). The intraclass correlation coefficient (ICC) provides an estimate of how much of the variance is due to grouping structure – the higher the ICC, the more homogenous units are and the greater variability there is between units (Heck et al, 2014). The ICC in the current sample was 0.77 (77%,  $p < .001$ ) thus indicating that a MLM approach is required because the higher-level grouping meaningfully affects the estimates.

In the next step of the analysis we introduced parent gender as a fixed factor in the model. Parent gender was not significantly associated with relationship satisfaction ( $p = .571$ ). Further potentially confounding socio-demographic indicators (family SEP, number of children in the family) were then modelled. Neither family SEP ( $p = .118$ ) nor number of children in the family ( $p = .759$ ) were significantly related to relationship satisfaction.

In the final step of the analysis we entered parent gender, family SEP, number of children in the family, parent depression, and the behavioural and emotional problems of the child with ASD and the sibling. In addition, interaction terms between depression and gender were included to explore whether a depression–relationship satisfaction association varied for mothers and fathers.

Models were fitted twice, once using the raw scores on dyadic adjustment and a second time using standardised DAS scores so as to obtain y-standardised coefficients for the predictors. The results of the final model with raw and y-

standardised coefficients are shown in Table 2.5. The addition of all predictors in the final model was associated with a change in the DAS score variance accounting for 9.1% within couples and 10.8% between couples.

### **Final Model**

In the final model, parental gender was not significantly associated with relationship satisfaction ( $p=.293$ ), with mothers and fathers reporting similar levels of relationship satisfaction. Level 2 variables, family SEP ( $p=.795$ ), number of children in family ( $p=.933$ ), and sibling behaviour problems ( $p=.773$ ), were also not significantly related to relationship satisfaction.

At Level 1, parental depression was significantly and negatively associated with relationship satisfaction ( $p=.007$ ), with elevated levels of depression related to lower levels of relationship satisfaction. However, the magnitude of this association was small:  $y$ -standardised coefficients indicate that as depression increases, relationship satisfaction scores decrease by .037 standard deviations. There was no evidence that this association was moderated by parent gender as the Gender x Depression interaction was not significant ( $p=.166$ ).

Behaviour problems of the child with ASD (Level 2) were significantly related to relationship satisfaction ( $p=.046$ ) with relationship satisfaction decreasing as the level of child behaviour problems increased. This was a small effect: as child behaviour problems increased, relationship satisfaction scores decreased by .019 standard deviations.

*Table 2.5 Final multilevel model results; raw and y-standardised coefficients of all fixed variables*

Variable	Raw coefficients	SE	y-standardised coefficients
Parent gender	-.401	.38	-.066
Family SEP	.141	.54	.023
Number of children in family	.056	.67	.009
Parental depression	-.228**	.08	-.037**
SDQ Child with autism	-.117*	.05	-.019*
SDQ Sibling	-.020	.07	-.003
Parent Gender x Depression	-.153	.11	-.025

*Note. SEP, Socioeconomic Position; SDQ, Strengths and Difficulties Questionnaire*

\* $p < .05$  \*\*  $p < .01$



## Discussion

The current study used a multilevel approach to explore whether mothers and fathers of children with ASD report different levels of relationship satisfaction and some of the factors that may be associated with this outcome. No difference in the way that mothers and fathers reported on their relationship was found, supporting other studies that have also found no gender differences on the DAS for parents of children with ASD (Lee, 2009; Gau et al, 2012). The finding suggests that mothers and fathers appear to experience similar levels of relationship satisfaction, rejecting the gender narrative. The absence of gender differences may reflect systems thinking in that parents have shared experience of child and family factors, and there may be an element of ‘crossover’ whereby one partner’s level of satisfaction transfers to the other partner in the household (Gerstein et al., 2009). Findings showed that the proportion of mothers and fathers categorised as being in the ‘distressed’ range on the DAS-7 was 44.6% (44.6% mothers, 44.5% fathers). This is a higher proportion than other studies that have used the DAS-7 among mothers of children with ASD (26%) (Benson & Kersh, 2011) and the full 32-item DAS in the general population (21% women, 22% men) (South, Krueger, & Lacono, 2009).

The study found an association between depression and relationship satisfaction scores. These findings echo other studies that have reported a significant and negative association between depression and relationship satisfaction in parents of children with ASD (Benson & Kersh, 2011; Shtayermman, 2013; Sim et al., 2016; Weitlauf et al, 2014). This association may not be unique to families of ASD as it has also been robustly reported in population-based studies (Fincham, Beach, Harold, & Osborne, 1997). It may be more likely to be reported in families raising a child with ASD because parents typically report elevated levels of depression compared to other groups (Abbeduto et al., 2004; Blacher & McIntyre, 2006; Gau et al., 2012; Lai et al., 2015), however in our sample the majority of parents reported depressive symptoms in the normal range.

Previous studies have found gender differences in depression levels with mothers of children with ASD reporting higher levels of depression (Jones et al., 2013; Lee et al., 2009). This was also found within the present study with mothers

reporting significantly higher depressive symptoms than fathers. However, our findings show that gender did not moderate the depression-relationship satisfaction relationship. The finding suggests that the association between mental health (depression in this case) and relationship satisfaction is experienced in a similar way by both mothers and fathers.

A negative association between child behaviour problems and relationship satisfaction in parents of children with ASD was found within the present study and contributes to the existing literature which has also reported this association (Benson et al., 2011; Hartley et al., 2012; Robinson & Neece, 2015; Sikora et al., 2013). FST refers to the importance of maintaining couple and parental boundaries for well-functioning subsystems, however these boundaries appear to be tested when raising a child with challenging behaviour. This association appears to be specific to the child with ASD given that this study found no evidence that sibling behaviour problems were related to parent relationship satisfaction. Parents may be more confident in dealing with the behaviour problems of a typically developing child producing a lesser impact on their partner role.

Family SEP was not found to be a statistically significant correlate of relationship satisfaction, once all other factors were accounted for. This finding contrasts previous work which has found an association between deprivation and relationship satisfaction in families of children with ASD (Benson et al., 2011; Hartley et al., 2012). Our analysis was largely exploratory and looked more broadly at SEP rather than deprivation, and the lowest SEP category included very few families ( $n=7$ ), suggesting limited variability of SEP scores. In contrast to previous research (Harper et al., 2013), no association was found between DAS scores and the number of children in the family, indicating that added emotional and financial pressures associated with having more children in the household, may not be related to how parents in this sample report on their relationship. However, it is important to note that unlike the study by Harper and colleagues, our study required families to have at least two children to participate.

The study has provided an initial insight into some of the factors associated with relationship satisfaction in families of children with ASD, however further

research is needed to expand upon the range of parent, child and family factors measured in the current study.

It is a challenge to study relationship satisfaction as a cross-sectional analysis, as relationships vary over time. The wide age range of the target children and parents may mean that families involved in the study were at different stages of the lifecycle and thus may be experiencing different stresses. It is also important to note that the DAS-7 omits other important relationship aspects such as decision-making, affection and sexual relations. It is also not certain whether mothers and fathers did indeed complete their questionnaires separately. Furthermore, studies like this one inevitably focus on relationship ‘survivors’- the 75% of parents of children with ASD that have remained together or are not experiencing significant relationship problems; therefore exploring co-parenting in future studies would include more parents.

The current study has furthered our understanding of relationship satisfaction in parents of children with ASD with a large dataset of 146 mother-father couples using an MLM approach. The study was designed to begin to provide evidence towards a theoretical model specific to the relationship satisfaction of parents of children with ASD and emphasised the importance of systemic thinking when exploring the family relationships of parents of children with a disability. With mothers and fathers reporting in the same way it suggests that the influence of family context on how parents report about themselves and their family could be greater than previously predicted. The influence of parent-level (depression) and child-level (child behaviour problems) variables on dyadic measures (relationship satisfaction) reinforce the importance of exploring factors and their interactions on a range of levels.

These findings have a number of practical implications, namely that the impact of raising a child with a disability should extend to explore the effect on family subsystems that do not just include children. Fathers and partners are just as likely to be affected as mothers in a positive or negative way by raising a child with a disability and professionals should continue to recognise the value of supporting all family members in relation to their family unit. The importance of supporting

parents of children with ASD to manage challenging behaviour is also clear given the negative outcomes it can have for the parent relationship. The negative association between individual levels of depression and relationship satisfaction points towards the need for services to be continually aware of the impact of such mental health difficulties on the personal relationships of parents of children with ASD, in order to provide timely support and advice.

**Chapter 3: The Psychological Wellbeing of Fathers with and without a Child  
with Intellectual Disability (ID): A Population-Based Study**

### Abstract

Few studies have explored the wellbeing of fathers despite the significant role that they play in their children's lives. The present study compared the psychological wellbeing of fathers with and without a child with Intellectual Disability (ID) using secondary data from the third wave of the Millennium Cohort Study (MCS), a UK population-representative longitudinal cohort study. Two-hundred and fifty-six fathers of a child identified as having ID, and 10,187 fathers without a child with ID, were compared on measures of individual wellbeing (life satisfaction, work-life balance, and general health) and parenting (parenting competency and parent-child closeness). Regression analyses then explored whether the presence of a child with ID was still a significant predictor of paternal wellbeing when controlling for a number of variables identified as significant correlates of wellbeing. Initial comparisons showed that fathers of children with ID reported lower levels of life satisfaction, and were more likely to report poor levels of general health, compared to fathers of children without ID. However regression analyses revealed that raising a child with ID was not a significant predictor of any of the paternal outcomes when controlling for a number of parent, child, and household variables. Raising a child with behavioural and emotional problems, and living in poverty, were found to be two significant predictors of paternal wellbeing, which has important implications for theory and practical support for fathers. Future research might begin to focus on the role of a variety of paternal wellbeing variables for the longer term developmental outcomes for children with and without ID.

**Key words:** Fathers, intellectual disability, wellbeing, population-representative

## **Introduction**

Theoretical frameworks such as Family Systems Theory (FST) recognise that fathers are an integral part of the family unit (Seligman & Darling, 2007). The birth and care of a child with a disability is understood to affect every member of the family, including the father, yet few studies have explored fathers' wellbeing in light of this knowledge (Braunstein, Peniston, Perelman, & Cassano, 2013; MacDonald & Hastings, 2010). Parental wellbeing is also considered to be a key factor in determining child outcomes. Developmental Systems Theory (Guralnick, 2005) describes how parental stress can be a risk factor for child development, indicating that paternal outcomes should be studied not only for their relevance to fathers, but also their children.

There have been relatively few studies that have conducted comparisons between fathers of children with ID and those who have typically developing (TD) children, with fathers' wellbeing often being compared instead to that of mothers (Olsson & Hwang, 2001). Research in this area has tended to explore more negatively-focused outcomes such as depression and stress. Findings suggest that fathers of children with ID are at heightened risk of experiencing mental health difficulties: Oelofsen and Richardson's (2006) study of 59 families of pre-school children with developmental disabilities (DD) and 45 families of pre-schoolers without DD, found that 67% of fathers of children with DD reported parenting stress levels within the clinical range compared to 10% of fathers of typically developing children. In another study by Giallo et al. (2015) in Australia that included 315 fathers of children with ID aged 3-15 years, fathers reported significantly more symptoms of depression, anxiety and stress on the Depression, Anxiety and Stress Scale (DASS) when compared to Australian normative data.

It is important that an exploration of paternal wellbeing does not only focus on symptoms of psychological distress but also other dimensions of wellbeing. As defined by the World Health Organization, mental health and wellbeing is a "state of complete physical, mental and social wellbeing and not merely the absence of disease or infirmity" (World Health Organization, 2014, para.2). Previous research has tended to focus more narrowly on the presence or absence of symptoms of

psychiatric disorder and not the other dimensions of mental health and wellbeing. In the present research, one aim was to extend the analysis to paternal life satisfaction, work-life balance, parenting competence, and relationship with their child with ID.

Subjective wellbeing and, in particular, life satisfaction of fathers raising a child with ID has received little attention within the literature. However, fathers of children with ID may be more likely to face additional challenges which have an impact on their outlook on life. Some, albeit limited, evidence on life satisfaction suggests that fathers of children with ID do report lower levels of life satisfaction. Darling, Senatore, and Strachan (2012) compared the life satisfaction of 85 fathers of children with disabilities (the most common disability being Attention Deficit Disorder, 36.5%) and 121 fathers of children with typically developing children using the five-item Satisfaction with Life Scale (SWLS; Diener, Emmons, Larsen, & Griffin, 1985). They found that fathers of children without a disability reported greater levels of life satisfaction than fathers of children with a disability.

The WHO definition of mental health and wellbeing also includes an individual being able to cope with the normal stresses of life and work productively and fruitfully (WHO, 2014, para. 1). Achieving a balance between work and family life may be more difficult to achieve when raising a child with a disability due to the additional considerations involved in child rearing. Fathers are more likely to be the main breadwinner (Gray, 2003) and so exploring their work-family life balance is important in relation to paternal wellbeing. Parenting and parent-child relationships are also considered by Developmental Systems Theory to mediate the impact of parental stress on child outcomes (Guralnick, 1997). Therefore, father-child relationships are important to explore in relation to the wellbeing of fathers and their children. Raising a child with a disability may pose additional challenges which impact upon fathers' parenting competence and the father-child relationship. In addition to narrow outcome measurement, a limitation of much of the existing research has also been that the studies have not explored evidence based on a large and representative sample of fathers. In Australia, the Longitudinal Study of Australian Children (LSAC) has been used to explore the risk of mental health problems for fathers of typically developing children aged 0-5 years (Giallo et al., 2012), and factors associated with trajectories of psychological distress in the early



parenting period (Giallo, D'Esposito, Cooklin, Christensen, & Nicholson, 2014). In the UK, national population representative surveys such as the Millennium Cohort Study (MCS) have been used to study the psychological wellbeing of mothers of children with ID or autism (Hatton & Emerson, 2009; Totsika et al., 2011a; Totsika et al., 2011b). Work by Emerson et al. (2010) has used MCS data to compare the presence of psychological distress in mothers and fathers with and without a child with early cognitive delay at age 3 (MCS Wave 2) and age 5 (Wave 3), however more population-based data on fathers of children with ID are needed.

It is clear that studying paternal wellbeing is of both theoretical and practical importance. Paternal wellbeing and parenting competency and closeness have the potential to impact upon other members of the family system, in particular children. To date, the MCS has not been used to compare fathers of children with and without ID on these dimensions (and population-based data are rare in any case), therefore such an analysis was warranted.

An additional question that it is important to explore is what factors may be associated with wellbeing in fathers. Existing research has explored a range of child and demographic variables, including the presence of a child with a disability in the family (see earlier), the behaviour problems of the child, child age, and family adversity. Increased child behavioural and emotional problems has been found to be associated with lower levels of paternal wellbeing including: psychological stress in fathers of toddlers with Pervasive Developmental Disorders (PDD) (Herring et al., 2006), depression and anxiety in fathers of children with ID (Cohen, Zeedyk, Tipton, Rodas, & Blacher, 2016; Giallo et al., 2015), and stress in fathers of children with Down syndrome (Ricci & Hodapp, 2003) and ASD (Brobst et al., 2009; Davis & Carter, 2008). Specific child characteristics such as gender may also be associated with fathers' wellbeing. In existing research, increased depressive symptoms have been associated with having a male child with ID (Trute, 1995), and increased paternal stress with having a female child with ID (Ricci & Hodapp, 2003) and autism (Rivard, Terroux, Parent-Boursier, & Mercier, 2014). The age of the child is also an important variable to consider given that there are likely to be different stressors at various stages of a child's life which can impact upon wellbeing. Evidence exploring the effect of child age on maternal wellbeing varies according to

the outcome: a meta-analysis of studies of maternal depression with and without a child with ID by Singer et al. (2006) indicated that maternal levels of depression improve with child age, with parents of adults having lower depression than parents of children in early and middle childhood. Conversely, a study that explored both positive perceptions and levels of anxiety and depression in mothers of children with ID aged 1-19 years, found no correlations between parental outcomes and the age of the child with ID (Vilaseca et al., 2013). It is clear from current evidence on mothers that it is important to explore associations between child characteristics such as gender and age and paternal wellbeing. Conducting a cross-sectional analysis focusing on a sample of children the same chronological age (e.g., at age 5 years in MCS) allows for a more targeted exploration of the variables associated with parental wellbeing at one particular time point and helps to control for developmental effects (Totsika et al., 2011a). Furthermore, at age 5 differences between children with and without ID may have become more evident, which could subsequently have an impact on paternal wellbeing.

There are also a range of environmental factors which can put families at risk of poorer outcomes, with one of the most significant being income poverty. The Developmental Systems Theory postulates that living in poverty and low levels of parental education are factors strongly associated with ‘non-optimal’ levels of child development (Guralnick, 1997). Families of children with ID are at increased risk of economic disadvantage compared to families of TD children (Emerson, 2003). Studies by Emerson and colleagues have repeatedly found an association between socioeconomic deprivation and wellbeing: a 2008 study found that 50% of the elevated risk of distress in mothers of young children at risk of a disability was accounted for by increased rates of poverty (Emerson & Llewellyn, 2008). A later study in 2010 using MCS data at Wave 2 (age 3) and Wave 3 (age 5) also found that socioeconomic deprivation accounted for differences in psychological distress between parents of children with and without early cognitive delay (Emerson et al., 2010). While a study by Giallo et al. (2015) did not find an association between socioeconomic status and symptoms of depression, anxiety and stress in fathers of children with ID, this could be attributed to the use of a measure of area-level of deprivation as opposed to individual level deprivation measures used in studies by Emerson and colleagues. More recent findings have also suggested that grouping

individual-level deprivation indicators in a composite may mask the different associations between wellbeing and socioeconomic indicators. For example, Totsika, Hastings and Vaagenas (2016) found that poverty was associated with reduced wellbeing among informal carers of people with ID, whereas having no educational qualifications was associated with improved wellbeing (e.g. less impact on personal life). These findings highlight the importance of selecting individual indicators of socioeconomic position to examine their separate associations with paternal wellbeing. The current analysis therefore included paternal education, employment status, and income poverty (above or below the 60% national equivalised median income).

The current study drew on UK population-representative data to compare the wellbeing of fathers of children with ID with fathers whose children do not have ID on measures of paternal wellbeing including life satisfaction, work-family balance, and general health, and measures of parenting competence and father-child closeness. The second aim was to explore whether the presence of a child with ID in the family was a significant predictor of paternal wellbeing when controlling for a number of variables identified as significant correlates of wellbeing. These included father variables such as age, employment, education and type of residency; child gender and parent-reported child behaviour problems; and family factors such as income poverty, and number of children in the household.

## Method

This study used data from Wave 3 of the Millennium Cohort Study (MCS3) when the cohort child was 5 years old (Centre for Longitudinal Studies, 2017). The MCS is a longitudinal birth cohort study tracking the lives of approximately 19,000 British children who were born in the UK in 2000-2001 (see [www.cls.ioe.ac.uk](http://www.cls.ioe.ac.uk)). Families were randomly selected using the Child Benefit register, which at the time of the study design was a non means-tested welfare benefit available to all UK children and with near universal coverage. Participants were drawn from 398 randomly selected electoral wards in the UK. Sampling was geographically clustered and disproportionately stratified using the Child Poverty Index (CPI) to ensure that children in all 4 countries (England, Wales, Scotland, Northern Ireland) from disadvantaged and ethnic minority backgrounds were adequately represented (Plewis, 2007). The decision to focus on the third wave of the MCS in the current study was based on child and father data available within this wave of data collection and because it may be that we begin to see differences in children at age five which subsequently have implications for paternal wellbeing.

## Participants

The analysis includes main and partner respondents in MCS3 who were fathers of the cohort child ( $N=10443$ ) (including biological, adoptive, step, and foster fathers). In the majority of cases (96.2%) the father was the partner respondent. Demographic information for fathers with and without a child with ID and comparison tests are presented in Table 3.1.

*Table 3.1 Comparison of demographic variables for fathers of children with and without ID (weighted)*

	Fathers of children without ID	Fathers of children with ID	
Variables	Mean(SD)	Mean(SD)	<i>t</i>
Father's age	36.4 (6.85)	37.3 (6.26)	1.57
Number of children in the home	2.3 (0.9)	2.9 (1.3)	-7.81*
Variables	%	%	$\chi^2$
Fathering type			
Biological father	95.6	92.6	71.9*
Adoptive father	0.2	-	
Stepfather	4.2	7.4	
Foster father	-	-	
Relationship between parent/carers in the household			
Married	77.9	63.1	29.5*
Cohabiting	21.3	35.2	
Not applicable (inc. single, separated, divorced, widowed).	0.8	1.7	
Ethnic group			
White	90.0	82.1	82.0*
Mixed	0.7	-	
Indian	2.3	0.9	
Pakistani and Bangladeshi	3.7	14.9	
Black or Black British	1.7	2.1	
Other	1.6	-	
Employment status			
In work	91.4	78.4	47.7*
Not in work	8.6	21.6	
Educational level			

	Fathers of children without ID	Fathers of children with ID	
Degree level or above	41.5	18.8	45.4*
Below degree level	58.5	81.2	
Father resident in the child's home			
Full time	99.1	97.9	3.87*
Part time	0.9	2.1	
OECD poverty median indicator			
Above 60% median	81.3	53.4	114.0*
Below 60% median	18.7	46.6	

Note. SD: standard deviation

\* $p < .05$

Fathers were similar in age (fathers without ID:  $M=36.4$  years,  $SD=6.85$ , Range =16-77; fathers with a child with ID:  $M= 37.3$  years,  $SD=6.26$ , Range =21-65). Of the 12 fathers of children without ID who were between 16- 20 years old (and thus would have been age 15 or younger when the cohort child was born), 11 were stepfathers. Both groups of fathers had an average of two children living in the household (Non-; ID=2.3,  $SD=0.9$ , Range=1-9; ID=2.9,  $SD=1.3$ , Range =1-8). The majority of fathers without a child with ID were biological fathers (95.6%), with the remaining stepfathers (4.2%) and adoptive fathers (0.2%). The majority of fathers of children with ID were biological fathers (92.6%), with the remaining stepfathers (7.4%). There was a statistically significant difference between the two groups on this variable ( $\chi^2(1) = 71.9, p<.001$ ). The majority of fathers without a child with ID were married to the other parent or carer in the household (77.9%), with the remaining cohabiting (21.3%) or “not applicable” (0.8%). Sixty-three percent of fathers of children with ID were married, with 35.2% cohabiting, and 1.7% selecting “not applicable”. There were statistically significant differences between the two groups ( $\chi^2(2) = 29.5, p<.001$ ). The majority of fathers of children without a child with ID described their ethnicity as White (90%). Eighty-two percent of fathers of children with ID described their ethnicity as White, with the remaining describing their ethnicity as Pakistani and Bangladeshi (14.9%), Black or Black British (2.1%) or Indian (0.9%). There were statistically significant differences between the two

groups ( $\chi^2(5) = 82.0, p < .001$ ). Ninety-one percent of fathers of children without ID were in employment at the time of the research, compared to 78.4% of fathers of children with ID, which was a statistically significant difference ( $\chi^2(1) = 47.7, p < .001$ ). Ninety-nine percent of fathers without a child with ID and 97.9% of fathers with a child with ID were full time residents in the child's home which was a statistically significant difference ( $\chi^2(2) = 3.87, p = .049$ ). Fifty-eight percent of fathers without a child with ID, and 81.2% of fathers of children with ID, had an educational qualification below degree level, which was a statistically significant difference ( $\chi^2(1) = 45.4, p < .001$ ). Eighteen percent of fathers of children without ID were in income poverty (below the 60% median poverty indicator) compared to 46.6% of fathers of children with ID. There was a statistically significant difference between the two groups ( $\chi^2(1) = 114.0, p < .001$ ).

The gender of the cohort child was equally split with 5324 (51%) males and 5119 (49%) females. To determine the presence of an intellectual disability a grouping variable created in a study by Totsika, Hastings, Emerson, & Hatton (2018) where ID was anchored at age 7 of the MCS (MCS4) was adopted. Age 7 was deemed by the study to be the best time for ID identification, as in the UK children have been in formal schooling for two years and there is evidence from prevalence studies to suggest that most children are identified as having ID at school-going age (Maulik et al., 2011). A principal components analysis (PCA) was conducted on age-standardised scores on two subscales of the British Ability Scales Second Edition (BAS II; Elliott, Smith & McCulloch, 1996): pattern construction and word reading, and a mathematics test (NFER Progress in Maths). This confirmed the presence of an underlying factor representing the child's general cognitive ability (named 'g') which accounted for 63% of the total variance. ID was defined as a g score equal or lower than two standard deviations below the mean ( $\leq 70$ ). Where children could not be identified as having ID at age 7 due to missing data, a PCA was conducted based on similar cognitive assessment data provided at age 5 (MCS3), 3 (MCS2), or parent and teacher reported information at age 7 about ID if cognitive assessment data was unavailable at these time points.

## Measures

In MCS3, a range of parent data was gathered using Computer Assisted Personal Interviewing (CAPI) and Computer Assisted Self-Interviewing (CASI). Paternal life satisfaction was measured using a single item which asked fathers to rate “how satisfied they were with the way their life had turned out so far” on a scale of 0 (*extremely unsatisfied*) to 10 (*completely satisfied*). This life satisfaction item is a subjective wellbeing measure which been used in large population-representative studies such as the MCS and national wellbeing surveys by the Office for National Statistics (ONS). Scores can be categorised as very low (0-4), medium (5-6), high (7-8), and very high (9-10) (Office for National Statistics, 2017). Paternal work-family balance was measured using a single item measure which asked fathers to rate “their satisfaction with work/family balance” on a scale of 1 (*very satisfied*) to 5 (*very dissatisfied*).

Fathers’ general health was measured using a single item measure which asked fathers to rate their overall health on a Five-point scale from 0 (*poor*) to 5 (*excellent*). The single item self-rated health measure is one of the most commonly used measures of global health status (Krause, 1994), which has been reliably used within clinical and research contexts (Bombak, 2013). Based on the distribution of this measure scores were dichotomized into two groups: 0=poor health (scores of 2 or lower) and 1=good health (scores of 3 or higher).

Parenting competence was measured using a single item measure which asked fathers to rate their parenting competence on a Five-point scale from 1 (*not very good at being a parent*) to 5 (*a very good parent*). Scores were dichotomised: 0=low level of parenting competence (scores of 1 and 2) and 1=high level of parenting competence (scores of 3 or higher). Parent-child closeness was measured using a single item measure which asked fathers to rate their relationship with their child on a Four-point scale from 1 (*not very close*) to 4 (*extremely close*). Scores were dichotomised into two groups: 0=not close (scores of 1 and 2) and 1=close (scores of 3 and 4).



The Strengths and Difficulties Questionnaire (SDQ; Goodman, 1997) was completed by primary caregivers for all the target cohort children as a measure of behavioural and emotional problems of their children (see Appendix R, p. 278). Primary caregivers were mostly mothers, however the current study also included SDQs completed by grandmothers. The 25-item scale generates scores in four problem domains: emotional symptoms, conduct problems, hyperactivity, and peer problems; and a pro-social behaviour domain. Caregivers indicate how likely each statement (e.g. “Often unhappy, downhearted or tearful”) applies to the child on a Three-point scale: *Not true*, *somewhat true*, *very true*, based on their child’s behaviour over the past six months. The SDQ is a reliable measure of psychological adjustment which has been used in research with typically developing (Goodman, 2001) and ID samples (Totsika et al., 2011a, Totsika et al., 2011b). The total problem difficulties score was used in the present study, with a higher score indicative of greater behavioural and emotional problems. Internal consistency was very good (Cronbach’s  $\alpha$ : Children with ID: 0.97 and children without ID: 0.93).

### **Procedure and Analysis Approach**

Data for MCS3 were obtained from the UK Data Archive ([www.ukdataservice.ac.uk](http://www.ukdataservice.ac.uk)). The initial step was to identify the total number of fathers in the MCS3 dataset, including fathers who were biological, adoptive, step and foster fathers among those who had responded as a main or a partner respondent. The next stage was to identify how many of these fathers had a child with ID by combining father data with the child ID variable. In total, there were 10,187 fathers without a child with ID, and 256 fathers with a child with ID. Information was collected in the MCS on all children in the instance of multiple births; however the present study focused only on the first target child in multiple birth households to avoid statistical complications. Analyses were conducted in SPSS Statistics 24.0® using the Complex Samples Procedure. Because of the MCS sample design, weights for design, sampling and attrition were applied to all the analyses following the creation of a Complex Samples Plan (CS Plan) (Jones & Ketende, 2010).

## Results

### Comparing Fathers' Wellbeing Scores

Between group comparisons of life satisfaction and work-family balance scores were conducted by independent *t*-tests using the general linear complex samples procedure. Fathers of children with ID reported lower levels of life satisfaction ( $M=7.10$ ,  $SE=.198$ ) compared to fathers of typically developing children ( $M=7.58$ ,  $SE=.023$ ), and the difference was statistically significant ( $t(1) = 2.466$ ,  $p = .014$ ), albeit small in terms of effect size ( $d=0.17$ ) (see Table 3.2). There were no significant differences reported between fathers of children with ID ( $M=2.99$ ,  $SE=.123$ ) and fathers of typically developing children ( $M=2.93$ ,  $SE=.015$ ),  $t(1) = -.445$ ,  $p = .656$ ) on the work-family balance outcome (Table 3.2).

*Table 3.2 T-Test results for fathers of children with and without ID on life satisfaction and work-family balance (Complex Samples)*

	Fathers of children without ID		Fathers of children with ID		<i>t</i>	Cohen's <i>d</i>
	M	SE	M	SE		
Life satisfaction	7.58	.023	7.10	.198	2.466*	0.17
Work-family balance	2.93	.015	2.99	.123	-.445	0.03

*Note.* \* $p < .05$

Chi square tests were conducted for the general health, parenting competency, and parent-child closeness measures using the complex samples procedure. On the general health measure there was a significant difference between the two groups of fathers, with fathers of children without a child with ID more likely to report good rather than poor levels of general health  $X^2(1, N=10431) = 15.228$ ,  $p < .001$ ) (Non-ID fathers=88.9%, ID fathers = 79.6%) (Table 3.3). There were no significant differences between the two groups of fathers on the parenting

competence  $X^2(1, N=9905) = .694, p = .405$  and parent-child closeness measures  $X^2(1, N=9944) = .847, p = .358$  (Table 3.3).

*Table 3.3 Chi Square results for fathers of children with and without ID on general health, parenting competency and parent-child closeness measures (Complex Samples)*

		Fathers of children without ID		Fathers of children with ID		
		%	Adjusted residual	%	Adjusted residual	<i>Adjusted F<sup>a</sup></i>
General health	Good	88.9	3.0	79.6	-3.0	15.2***
	Poor	11.1	-3.0	20.4	3.0	
Parenting competency	High	96.0	.741	94.9	-.741	.694
	Low	4.0	-.741	5.1	.741	
Parent-child closeness	High	90.1	.846	87.9	-.846	.358
	Low	9.9	-.846	12.1	.846	

*Note.* <sup>a</sup> Adjusted F (for categorical variables)=F statistic for design-based Pearson chi-square that is converted to F test to account for the MCS sampling design

\*\*\*  $p < .001$

### **Factors Associated with Father Wellbeing**

Regression models using complex samples procedure were conducted separately for life satisfaction, work-family balance, general health, parenting competence, and parent-child closeness outcomes to analyse the main effects of the following variables: whether the father has a child with ID, child gender, child SDQ score, father age, father education, father employment, father residency, income poverty, and number of the children in the household. Linear regression models were used for the life satisfaction and work-life balance outcomes and logistic regression for the general health, parenting competence, and parent-child closeness outcomes. As it is not possible to compute a single  $R^2$  statistic using Complex Samples Procedure, the Cox and Snell pseudo  $R^2$  was used instead.

As shown by the unstandardized coefficients in Table 3.4, being in employment ( $B = .524, p < .001$ ) and living in a household above the 60% poverty median ( $B = .366, p < .001$ ) were positively associated with life satisfaction. Child SDQ scores were negatively associated with life satisfaction ( $B = -.021, p < .001$ ), as child behavioural and emotional problems increased, life satisfaction scores decreased. Increasing numbers of children in the household was positively associated with life satisfaction scores ( $B = .067, p < .01$ ). Child ID ( $B = .170, n.s$ ), child gender ( $B = .008, n.s$ ), father age ( $B = -.007, n.s$ ), father education level ( $B = -.043, n.s$ ), and father residency ( $B = -.401, n.s$ ), were not statistically significant. The overall model fit was pseudo  $R^2 = .022$ , a low level of explained variance.

Living in a household above the 60% poverty median ( $B = .187, p < .001$ ) and child behavioural and emotional problems ( $B = .007, p < .05$ ) were positively associated with work-family balance (Table 3.4). Father age ( $B = -.007, p < .01$ ) was negatively associated with work-family balance. Child ID ( $B = -.168, n.s$ ), child gender ( $B = .003, n.s$ ), father educational level ( $B = .016, n.s$ ), father residency ( $B = -.106, n.s$ ), and the number of children in the household ( $B = .018, n.s$ ), were not statistically significant (Table 3.4). Father employment was not included in the model because the measure was completed by working fathers only. The overall model fit was pseudo  $R^2 = .005$ , a low level of explained variance.

*Table 3.4 Linear regression models for life Satisfaction and work-family balance (Complex Samples)*

	Life satisfaction		Work-life balance	
	Coeff. (SE)	95% CI	Coeff.(SE)	95% CI
Constant	6.894***(.307)	[6.292—7.497]	3.119***(.184)	[2.758—3.480]
Child with ID	.170(.224)	[-.271—.611]	-.168(.141)	[-.445—.109]
Child Gender	.008(.042)	[-.075—.091]	.003(.029)	[.053—.060]
Child SDQ	-.021***(.005)	[-.030— -.012]	.007*(.003)	[.001-.014]
Father age	-.007(.004)	[-.014—.001]	-.007**(.003)	[-.012—-.002]
Father education	-.043(.045)	[-.132—.045]	.016(.033)	[.048—.080]
Father employment	.524***(.118)	[.292—.755]	-	-
Father residency	-.401(.250)	[-.892—.090]	-.106(.178)	[-.456—.224]
Income poverty	.366***(.073)	[.222—.510]	.187(.051)***	[.087—.287]
Number of children in household	.067**(.004)	[.016—.118]	.018(.018)	[-0.17—.052]

\* $p < .05$ , \*\* $p < .01$ , \*\*\* $p < .001$ .

Table 3.5 Logistic regression model for general health (Complex Samples)

	<i>B</i>	<i>SE</i>	<i>Wald</i>	<i>df</i>	<i>p</i>	<i>OR</i>	<i>OR 95% CI</i>
Constant	-2.506	.409	87.569	1	<.001		
Child with ID	-.035	.273	.017	1	.897	.965	[.564 – 1.651]
Child Gender	.009	.096	.010	1	.922	1.009	[.836 – 1.219]
Child SDQ score	.044	.008	29.188	1	<.001	1.045	[1.028 – 1.061]
Father age	.025	.007	11.377	1	.001	1.025	[1.011 – 1.041]
Father education	.559	.106	27.772	1	<.001	1.748	[1.419 – 2.153]
Father employment	-1.253	.118	113.053	1	<.001	.286	[.227 – .360]
Father residency	-.397	.424	.877	1	.350	.672	[.292 – 1.548]
Income poverty	-.410	.111	13.647	1	<.001	.664	[.534 – .826]
Number of children in household	.068	.044	2.416	1	.121	1.071	[.982 – 1.167]

*Note.* *B* estimated value of the regression coefficient, *SE* Standard error, *Wald* Wald statistic, *df* degrees of freedom, *p* level of significance, *OR* Odds Ratio, *OR 95% CI* Odds ratio with a 95% confidence interval, *SDQ* Strengths and Difficulties Questionnaire

Logistic regression models were conducted for the general health, parenting competence, and parent-child closeness outcomes. Child behavioural and emotional problems and father age were significantly associated with general health (Table 3.5). Fathers of children with higher SDQ scores ( $OR=1.045, p<.001$ ) and older fathers ( $OR=1.025, p=.001$ ) were more likely to report poor levels of general health. A father's education status, employment status, and family income poverty level were also significantly associated with general health. Fathers who possessed a degree level qualification were more likely to report poor levels of general health ( $OR=1.748, p<.001$ ). Fathers in work ( $OR=.286, p<.001$ ) and not living in poverty ( $OR=.664, p<.001$ ) were less likely to report poor general health. Child ID ( $OR=.965, n.s$ ), child gender ( $OR=1.009, n.s$ ), father residency ( $OR=.672, n.s$ ), and number of children in the household ( $OR=1.071, n.s$ ), were not statistically significant. The overall model fit was pseudo  $R^2 = .047$ , a low level of explained variance.

Child SDQ score, father age, and father employment, were significantly associated with parenting competence (Table 3.6). Fathers of children with higher SDQ scores ( $OR=1.043, p=.001$ ) and older fathers ( $OR=1.025, p=.004$ ) were more likely to report low parenting competence. Fathers in work were less likely to report low parenting competence ( $OR=.584, p<.05$ ). Child ID ( $OR= 1.304, n.s$ ), child gender ( $OR= 1.198, n.s$ ), father education ( $OR= .966, n.s$ ), father residency ( $OR= 1.820, n.s$ ), income poverty ( $OR= .901, n.s$ ) and number of children in the household ( $OR= 1.060, n.s$ ) were not statistically significant (Table 3.6). The overall model fit was pseudo  $R^2 = .005$ , a low level of explained variance.

In the parent-child closeness model (Table 3.7), number of children in the household, child SDQ score, and income poverty were statistically significant. Fathers with more children in the household ( $OR= 1.194, p<.001$ ), and fathers of children with higher SDQ scores ( $OR= 1.069, p<.001$ ) were more likely to report a poor relationship with their child. Fathers who did not live in poverty ( $OR= .743, p=.009$ ) were less likely to report a poor relationship with their child. Child ID ( $OR= 1.417, n.s$ ), child gender ( $OR= 1.039, n.s$ ), father age ( $OR=.999, n.s$ ), father education ( $OR=.943, n.s$ ), father employment ( $OR= 1.450, n.s$ ), and father residency

( $OR=1.426$ , n.s) were not statistically significant (Table 3.7). The overall model fit was pseudo  $R^2 = .015$ , a low level of explained variance.



Table 3.6 Logistic regression model for parenting competency (Complex Samples)

	<i>B</i>	<i>SE</i>	<i>Wald</i>	<i>df</i>	<i>p</i>	<i>OR</i>	<i>OR 95% CI</i>
Constant	-.4327	.616	73.388	1	<.001		
Child with ID	.266	.360	.544	1	.461	1.304	[.642—2.649]
Child Gender	.180	.123	2.148	1	.144	1.198	[.940—1.526]
Child SDQ score	.042	.013	10.848	1	.001	1.043	[1.017—1.069]
Father age	.025	.009	8.175	1	.004	1.025	[1.008—1.043]
Father education	-.035	.124	.080	1	.778	.966	[.758—1.231]
Father employment	-.538	.272	3.098	1	.049	.584	[.342—.997]
Father residency	.599	.496	1.458	1	.228	1.820	[.686 —4.828]
Income poverty	-.105	.185	.320	1	.572	.901	[.626—1.296]
Number of children in household	.058	.068	.724	1	.395	1.060	[.927—1.213]

*Note.* *B* estimated value of the regression coefficient, *SE* Standard error, *Wald* Wald statistic, *df* degrees of freedom, *p* level of significance, *OR* Odds Ratio, *OR 95% CI* Odds ratio with a 95% confidence interval, *SDQ* *Strengths and Difficulties Questionnaire*

Table 3.7 Logistic regression model for parent-child closeness (Complex Samples)

	<i>B</i>	<i>SE</i>	<i>Wald</i>	<i>df</i>	<i>p</i>	<i>OR</i>	<i>OR 95% CI</i>
Constant	-3.523	.485	61.998	1	<.001		
Child with ID	.349	.279	1.567	1	.211	1.417	[.820—2.452]
Child Gender	.038	.080	.224	1	.636	1.039	[.887—1.216]
Child SDQ score	.067	.007	80.807	1	<.001	1.069	[1.054—1.085]
Father age	-.001	.007	.010	1	.921	.999	[.985—1.014]
Father education	-.058	.085	.477	1	.490	.943	[.799—1.114]
Father employment	.371	.190	3.840	1	.051	1.450	[.999—2.105]
Father residency	.355	.345	1.056	1	.305	1.426	[.723—2.813]
Income poverty	-.298	.114	6.815	1	.009	.743	[.593—.929]
Number of children in household	.178	.044	16.409	1	<.001	1.194	[1.096—1.302]

*Note.* *B* estimated value of the regression coefficient, *SE* Standard error, *Wald* Wald statistic, *df* degrees of freedom, *p* level of significance, *OR* Odds Ratio, *OR 95% CI* Odds ratio with a 95% confidence interval, *SDQ* Strengths and Difficulties Questionnaire

## Discussion

This study is among the first to compare multiple measures of wellbeing and parenting between fathers of children with and without a child with ID in a population-based sample. The findings suggest that the wellbeing of fathers is not associated with having a child with ID. While fathers did report lower levels of life satisfaction (supporting previous work by Darling et al., 2012) and poorer general health outcomes in the pairwise comparisons, raising a child with ID was not a significant predictor of any of the paternal outcomes (life satisfaction, work-family balance, general health, parenting competency, parent-child closeness) in the regression models, when controlling for a number of parent, child, and household variables. Previous work has indicated that there may be differences in wellbeing for fathers raising a child with ID (Darling et al, 2012; Emerson et al, 2010; Giallo et al., 2015; Oelofsen & Richardson, 2006), however this study suggests that there were no significant differences when exploring measures of subjective wellbeing and parenting with a large and nationally representative sample of fathers.

The current findings allow us to extend existing systems frameworks and start to develop a theory specific to father wellbeing. In contrast to population-based studies with mothers of children with ID (Totsika et al., 2011a) and early cognitive delay (Emerson et al., 2010), these results which showed that there were no differences between fathers of children with and without a child with ID indicate that wellbeing outcomes could, in theory, be different for mothers and fathers. One explanation could be the association with caregiving roles, which are still predominantly assumed by females. Theoretical models outside of the ID field such as social role theory state that roles within the family are still distributed according to gender, with women more likely than men to be the primary caretakers of their children (Eagly & Wood, 2016). Therefore, it could be that a mother's wellbeing is more likely to be affected by raising children, particularly when a child has a disability. On a practical level, it suggests that mothers and fathers may require different types of support: mothers may benefit from more help with caregiving responsibilities, whereas fathers may not due to this role distribution.

Identifying factors that contribute to fathers' psychological wellbeing allows us to better understand the support needs of fathers and how we can achieve positive outcomes. Child behavioural and emotional problems were found to be predictive of all of the wellbeing measures in this study. This association has been reported in other studies which have explored the association between child behaviour and psychological distress (Herring et al., 2006), depression, anxiety (Cohen et al., 2016; Giallo et al., 2015) and stress (Brobst et al., 2009; Ricci & Hodapp, 2003) of fathers of children with disabilities, and can now also be extended further to these paternal wellbeing and parenting outcomes. Given that both paternal wellbeing and parent-child relationships are suggested by the Developmental Systems Theory to impact upon a child's outcomes, it is an important factor to consider. Furthermore, the finding also highlights that we should be focussing more on supporting fathers who have a child with behaviour problems, rather than primarily on a child's diagnostic label (Cohen et al., 2016).

The finding that there are a range of socioeconomic indicators, in particular income and employment status, which can predict both fathers' wellbeing and parenting outcomes, highlights an existing complex issue related to individuals who are at risk of poverty. Studies on families of children at risk of disability and early cognitive delay have consistently reported an association between socioeconomic deprivation and wellbeing (Emerson & Llewellyn, 2008; Emerson et al., 2010), and this study demonstrates that this is an association which affects fathers irrespective of whether they have a child with ID. Fathers are still more likely than mothers to assume a full-time role in paid work (Eagly & Wood, 2016), and thus income and employment is likely to have a significant impact on their wellbeing. Given that environmental factors such as poverty are considered to be associated with poorer parental wellbeing and non-optimal child development (Guralnick, 2005), support and intervention needs to target those most at risk in order to promote positive outcomes for fathers and their families.

The current study has furthered our understanding of paternal wellbeing using a large representative sample of fathers, however further research will be necessary to continue to explore a broader range of factors related to paternal wellbeing. While the MCS collects a vast amount of information from caregivers, the

majority of this is collected from primary caregivers who are usually mothers. It is also important to remember that the paternal outcomes in this study were measured by self-report. This is particularly pertinent in relation to parenting outcomes, where observational studies of fathers' parenting and father-child relationships are needed. While cross-sectional analyses have allowed us to focus on factors specific to fathers of children at a particular stage of their lifecycle, it would be useful to investigate father wellbeing longitudinally with further waves of the MCS. It is also important to note that this analysis focused on fathers who lived in the same household as the child for all or part of the time and therefore findings cannot be generalised to fathers who were not living in the same household as the child at the time of data collection.

The study was conducted to understand whether there were any differences in wellbeing between fathers of children with and without ID and identify some of the factors associated with their wellbeing. For some time now there has been interest in the wellbeing of parents and the subsequent outcomes of their children, however this has often been based on research with mothers. It cannot be assumed that fathers have the same experiences as mothers, and therefore research which continues to focus on the outcomes of fathers would be productive for both the academic community and those working to support fathers and their families. Future research might fruitfully begin to focus on the role of a variety of paternal wellbeing variables for the longer term developmental outcomes for children with and without ID – beyond the cross-sectional and father outcome focus of the present research.

**Chapter 4: Family Relationships and their Associations with Perceptions of  
Family Functioning in Mothers of Children with Intellectual Disability (ID)**

### Abstract

Raising a child with a disability is proposed to have an impact on core subsystems in the family and the overall family unit. However, there has been very little ID research which has explored functioning at the subsystem level and its associations with broader family functioning. The present study explored whether mothers' reports of three dyadic relationships (mother-father, parent-child, child-sibling) were related to mothers' perceptions of family functioning. The research included 431 mothers of children with ID aged 4-15 years who took part in the Cerebra 1000 Families Study and provided data on their relationship in three subsystems and family functioning. A confirmatory factor analysis of latent family functioning was conducted and then structural equation models were fitted to examine the associations between relationship indicators and family functioning. The latent family functioning factor achieved a statistically good fit, with Affection (I am satisfied with the way my family expresses affection and responds to my emotions, such as anger, sorrow and love) having the strongest loading. The final structural model showed that partner relationship satisfaction, partner disagreement, child-parent conflict, and sibling relationship warmth accounted for the most variance in family functioning. Standardised regression weights showed that partner relationship satisfaction had the strongest positive association with family functioning. While this was an exploratory study our data show that functioning in dyadic relationships is associated with broader constructs of family functioning in families of children with ID. Specifically, our findings suggest that interventions which target the marital/partner subsystem could improve the overall functioning of the family unit.

**Key words:** Family functioning, family systems, intellectual disability, confirmatory factor analysis, structural equation modeling

## **Introduction**

According to Family Systems Theory (FST) the family is a unit that can comprise a number of subsystems, including the marital/couple, parental, and sibling subsystem (Cox & Paley, 1997). From this perspective, the family is viewed as an interconnected system where raising a child with a disability impacts on all family members in these subsystems (Gardiner & Iarocci, 2012; Seligman & Darling, 2007). Much of the family research in the ID field has explored the individual as the core unit of analysis, rather than the family (Hoffman et al., 2006). Some ID family research has focused on interactions within and between family subsystems, however far less research has focused on larger triadic or whole-family questions, perhaps due to the level of complexity involved in conceptualising and measuring outcomes or functioning at the broader family system level (Cox & Paley, 1997; Hayden et al., 1998). Different knowledge can be gained from exploring experience at the family rather than the individual or dyadic level (Zuna, Summers, Turnbull, Hu & Xu, 2010). Broadening the unit from the ‘individual’ to the ‘family’ not only recognises the impact on other members within the family system, but how other family members and subsystems may in turn influence the wellbeing of a family member with a disability (Rolland, 2012).

One family-level construct that has been explored by researchers in the disability field (Brown et al., 2006; Poston et al., 2003; Turnbull, Summers, Lee, & Kyzar, 2007), is Family Quality of Life (FQOL). FQOL has been defined as “conditions where the family’s needs are met, and family members enjoy their life together as a family and have the chance to do things which are important to them” (Park et al., 2003, p.368). However, empirical research on FQOL has focused primarily on the development of the FQOL construct and how to measure it (Gardiner & Iarocci, 2012; Hastings, 2016).

Another family-level construct that has received some research attention is family functioning, which arose from Family Systems Theory (Summers et al., 2005). Family functioning can be defined in a variety of ways, but most definitions comprise the extent to which members of the family unit communicate, build relationships, and manage daily life (Jellet, Wood, Giallo, & Seymour, 2015).



Family functioning has been found to be associated with parent and child outcomes in families with typically developing children. For example, using large-scale normative data of caregivers of typically developing children, Renzaho, Mellor, McCabe, and Powell (2011) found that after controlling for socioeconomic status (SES) and ethnicity, parents in poorly functioning families were at greater risk of psychological distress and had children with lower levels of prosocial behaviour and higher levels of behavioural difficulties.

The construct of functioning appears particularly pertinent to families raising a child with ID as they are likely to face additional challenges which have an impact on family interactions and processes (McConkey, Truesdale-Kennedy, Chang, Jarrah, & Shukri, 2008). Parenting a child with ID requires more extensive family involvement, often with maternal caregiving and family interactions focused on the needs of the child with ID – which can mean that the family may lack balance (Brown et al., 2006). Family functioning has been shown to be poorer in families of children with ID (Al-Krenawi, Graham, & Al Gharaibeh, 2011; Rani et al., 2018) and autism (Gau et al., 2012; Higgins, Bailey & Pearce, 2005; Pisula & Porębowicz-Dörsmann, 2017) when compared to families with typically developing children. In addition, family functioning has been found to be a predictor of caregivers' emotional health in families of adults with ID, with maladaptive family functioning related to poorer psychological outcomes (Magaña, Schwartz, Rubert, & Szapocznik, 2006). Understanding what contributes towards family functioning in families of children with ID, would inform the design and delivery of family-centred support (Hoffman et al., 2006). There is theoretical and empirical evidence to suggest that functioning in one family subsystem can influence functioning in another due to the interdependent and reciprocal nature of family relationships. For example, there is some linkage between marital relationships and parent-child relationships, and conflict can “spillover” from one subsystem into the other (Erel & Burman, 1995). Hartley et al. (2016a) found that there was an association between marital interactions and parenting stress in families of children with autism. However, no evidence in ID research has examined whether functioning at the subsystem level is associated with functioning at the broader family system level. For example, it could be that how a mother feels about her relationship with her partner, with her child, or

the relationship between children in their family, may have some bearing on how satisfied she feel with her family overall.

The couple subsystem is considered to be at the heart of the family system, with its stability having implications for others in the family unit (Seligman & Darling 2007). Feldman, Wentzel, Weinberger, & Munson (1990) argue that “the quality of the marriage can have a pervasive effect on family life as a whole as well as individual outcomes of its members” (pp.213-214). Marital (or partner) satisfaction has been found to be a predictor of family-level outcomes. Early work by Trute (1990) explored child and parent predictors of family adjustment in 88 families of children with developmental disabilities, reporting that overall family adjustment (as captured by the Family Assessment Measure III; Skinner, Steinhauer, & Santa-Barbara, 1983) was associated with specific aspects of marital adjustment (dyadic cohesion, and consensus). Trute suggested that strengthening the couple subsystem could assist in maintaining a stable family environment. In a study by Henderson and colleagues (2003) with 43 mothers of boys identified as having behavioural difficulties, there was a statistically significant relationship between family cohesion (subscale of the Family Environment Scale; Moos & Moos, 1981) and marital satisfaction: the lower the perceived levels of cohesion in the family, the greater the marital discord (Henderson, Sayger, & Horne, 2003). Correlations between couple negativity and subsequent negative family interactions (Kitzmann, 2000), and between maternal marital satisfaction and overall family functioning (Feldman et al., 1990) have also been found in studies within the general population. Thus, it is possible that the marital/partner system may have a significant bearing on how satisfied a mother feels with how her family functions overall.

The parenting subsystem may also be related to family functioning. Parent-child relationships can be affected when raising a child with ID (Hastings, 2016; Totsika, Hastings, Vagenas, & Emerson, 2014), and the quality of this relationship has been linked to the psychological wellbeing of mothers (Greenberg, Seltzer, Krauss, Chou, & Hong, 2004). However due to a lack of empirical evidence, it is not clear whether the parent-child relationship is associated with broader family-level outcomes such as family functioning. The presence of emotional and behavioural problems in the child is significantly associated with poorer family functioning in

families of children with ID (Herring et al., 2006; Jellett et al., 2015). Higher levels of child behavioural and emotional problems may reduce the quality of the parent-child relationship which may in turn affect parents' perceptions of family functioning.

The sibling subsystem is also an aspect of family functioning which requires further exploration. The sibling relationship is usually one of the longest lasting relationships an individual can have (Dunn, 2000), and the outcomes for siblings of children with a brother or sister with ID/autism have been the focus of a number of empirical studies (Mulroy, Robertson, Aiberti, Leonard & Bower, 2008; Neece, Blacher, & Baker, 2010; Petalas, Hastings, Nash, Lloyd & Dowey, 2009; Rossiter & Sharpe, 2001; Stoneman, 2005). However, Kramer and Bank (2005) state that "in comparison to other family relationships, such as parent-child and marital relations, the contributions of sibling relationships to individual development and family functioning have been given scant attention" (p.483). This conclusion is particularly relevant to ID family research, where sibling psychological functioning has been shown to affect outcomes for children with ID (Hastings, 2007; Hastings et al., 2014), but associations between sibling relationship quality and parents' perceptions of family functioning have not been examined.

The present study explored three family subsystems (marital, parental, and sibling relationships) and their associations with family functioning. Although, based on FST, we might expect associations between the quality of relationships in family subsystems and family functioning as a whole, the lack of current ID family research evidence meant that there were no specific hypotheses and the study was exploratory.

## Method

### Participants

Four-hundred and thirty-one mothers of children with ID who took part in the Cerebra 1000 Families Study hosted at the University of Warwick provided data on family functioning and their relationship in three subsystems: marital/partner, parent-child, and child-sibling. The majority were biological mothers ( $n=40$ , 93.5%) and lived with their spouse ( $n=368$ , 85.4%) (Table 4.1). Two hundred and nine mothers (48.5%) were educated to university degree level or higher, with 227 (52.7%) not in work at the time of the research. The majority of mothers described their ethnicity as White ( $n= 375$ ; 92.3%). The modal weekly household income reported by mothers was £800-£1000 (British pounds sterling: approximately \$1068-\$1335 US dollars): 122 (28.3%). Families had on average two children living in the household ( $M=2.52$ ,  $SD=0.83$ , Range= 1-7).

All children who were reported on in the survey were reported by their mother to have ID. Two-hundred and twenty-three (51.7%) children were reported to have a 'mild/moderate' ID, and 203 (47.1%) a 'severe/profound' ID. Just over half of the sample were also reported to have autism ( $n=222$ , 52.2%). Children were on average 9 years of age ( $SD=2.91$ , Range = 4-15 years) and most were male: 293 (68.0%).

Parents were asked to answer questions about any sibling between the ages of 4 and 15 years. If there was more than one sibling in this age range, they were asked to select the child closest in age to the child with ID. Two-hundred and twenty siblings were male (51.0%) and 207 (48.5%) were female. The majority of siblings were the same gender: 228 (52.9%). Siblings of the child with ID were on average 8 years of age ( $SD=3.21$ , Range = 4-15 years). Two-hundred and sixteen (50.1%) siblings were younger than the child with ID, 207 (48.0%) were older, and one pair were the same age (0.2%). The majority of siblings (74.0%) were not considered to have a longstanding illness, disability or infirmity.

*Table 4.1 Demographic profile of mothers (N=431) and their child with intellectual disability and a sibling*

Variables	%
Mother type	
Biological	93.5
Adoptive	6.0
Step	0.2
Foster	0.2
Marital status	
Married and living with spouse	85.4
Living with partner	14.6
Education level	
University degree level or above	48.5
Below degree level	46.4
Missing	4.6
Employment status	
In work	47.1
Not in work	52.7
Missing	0.2
Ethnic group	
White	92.3
Mixed	0.9
Indian	0.9
Pakistani and Bangladeshi	1.2
Black or Black British	0.9
Ethnic other	1.4
Missing	1.4
Weekly household income	
£200 or less	1.9
£201-400	16.9
£401-600	27.1
£601-800	22.3
£801-1000+	28.3
Missing	3.5

Variables	%
ID severity	
Mild/Moderate ID	51.7
Severe/Profound ID	47.1
Missing	1.2
Child gender	
Male	68.0
Female	31.8
Missing	0.2
Sibling gender	
Male	51.0
Female	48.5
Missing	0.9
Sibling illness/disability	
Yes	26.0
No	74.0
	Mean (SD)
Child age	9.06 (2.91)
Sibling age	8.97 (3.21)
Number of all children in the household	2.52 (0.83)
Number of all people in the household	4.64 (0.92)

Note. SD: Standard Deviation

## Maternal Measures

The Family APGAR scale (Smilkstein, 1978) was used to assess perceptions of family functioning as reported by mothers (see Appendix O, p. 275). The Family APGAR was designed to capture five components of family functioning: Adaptation, Partnership, Growth, Affection and Resolve. Sample items from the measure include: “I am satisfied that I can turn to my family for help when something is troubling me” (Adaptation), and “I am satisfied with the way my family expresses affection and responds to my emotions, such as anger, sorrow and love” (Affection). Mothers were asked to rate 5 items on a Three-point scale: *Almost Always* =2, *Some of the time* =1, *Hardly ever*=0. The measure is scored by summing the values for the

items for a total score that can range from 0 to 10. A higher score indicates a greater degree of satisfaction with family functioning. Scores can also be used to categorise families into dysfunctional (0-3), moderately dysfunctional (4-7) and highly functional (8-10). The Family APGAR questionnaire has been used previously in studies investigating family functioning in the general population (Gardner et al., 2001) and has more recently been used to study family functioning where there is a child with a disability (Rani et al., 2018). Internal consistency (Cronbach's  $\alpha$ ) in the present study was .87.

Two single item measures were used to capture the mothers' relationship with their spouse or partner. The first item is a global measure of relationship happiness. Mothers were asked to select "the number which best describes how happy or unhappy you are with your relationship, all things considered" on a scale of 1 (*very unhappy*) to 7 (*very happy*). The second item asked mothers to rate "how often do you and your [husband/wife/partner] disagree over issues related to your child?" on a scale of 1 (never) to 6 (more than once a day). Both these items have been used in waves of large UK cohort studies such as the Millennium Cohort Study (Johnson, 2012).

### **Child Measures**

The Child-Parent Relationship Scale Short Form (CPRS-SF) was used to measure the quality of the parent-child relationship (see Appendix P, p.276). The CPRS-SF is adapted from the Student-Teacher Relationship Scale (STRS; Pianta, 1992) and is a 15 item scale asking parents to rate their relationship with their child. Items are measured on a scale of 1 (*definitely does not apply*) to 5 (*definitely applies*). Item scores are summed to provide scores for two dimensions: closeness, and conflict. Seven items are summed for closeness, and eight items for conflict. Higher scores indicate greater closeness or conflict in the parent-child relationship. The measure has been used in studies of parents of children with ID (Totsika et al., 2014). Sample items include "I share an affectionate relationship with this child" (Closeness), and "This child and I always seem to be struggling with each other" (Conflict). Internal consistency (Cronbach's  $\alpha$ ) in the present study was .76 for Closeness, and .85 for Conflict.

A shortened version of the Sibling Relationship Questionnaire – revised (SRQ brief parent-version; Furman & Buhrmester, 1985) was used to assess mothers’ perceptions of sibling relationship quality (see Appendix Q, p.277). The SRQ brief version is a 39-item questionnaire which measures 16 dimensions of sibling relationship on four scales: warmth/closeness, relative status/power, conflict, and rivalry. In the present study participants completed a shortened version of the measure including 10 items that captured dimensions of warmth/closeness and conflict. Mothers completed the three subscales of the Warmth factor (Intimacy, Companionship, Affection), and two scales of the Conflict factor (Quarrelling and Antagonism). Each of the subscales consisted of two items. Mothers were asked to read the 10 items and rate how much they apply to the siblings on a Five-point scale from 1 (*hardly at all*) to 5 (*extremely much*). The brief parent-version of the SRQ has been used before to assess the relationship quality of children with autism and their siblings (Petalas et al., 2012). Sample items include “How much do the sibling and the child love each other?” (Affection), and “How much do the sibling and the child disagree and quarrel with each other?” (Quarrelling). Scores for Warmth and Conflict were derived by calculating a weighted mean score from 1-5 for each of these two relationship domains. Internal consistency (Cronbach’s  $\alpha$ ) in the present study was .74 for Warmth and .81 for Conflict.

## **Procedure**

The data used for the present analysis were part of a large survey of parents of children with ID aged 4 to 15 years in the UK known as ‘The Cerebra 1000 Families Study’. Following ethical approval from the National Health Service (NRES West Midlands, ref: 15/WM/0267) (see Appendix A, p. 150), information about the study was distributed via a number of third-sector organisations who support families of children with ID. Recruitment took place mostly online through social media and locally through advertising via local parent support groups. A primary caregiver was asked to complete an online or paper survey, followed by a telephone interview. A total of 1082 caregivers took part in the study. The present research focuses on the data from all 431 mothers who reported being married and living with their spouse or living with a partner, and provided data on a sibling in the



household (including reporting on sibling relationship quality with the child with ID), and also reported on their relationship with their child with ID.

## **Analysis**

Our overall research question was: are dimensions of dyadic relationships (family subsystems) in the family related to mothers' perceptions of family functioning? To address this research question, a series of Structural Equation Models (SEM) were fitted in AMOS 24®. In comparison to other statistical analysis techniques such as regression, SEM is capable of testing more sophisticated theory (Schumacker & Lomax, 2012). It can test how variables define constructs, simultaneously test how constructs are related to each other, and explicitly take into account measurement error when analysing data (Schumacker & Lomax, 2012). Models were estimated using Maximum Likelihood (ML), a consistent and unbiased approach to parameter estimation commonly employed in SEM (Hair, Black, Babin, Anderson, & Tatham, 2006). Full Information Maximum Likelihood (FIML) was used to account for missing values in the sample ( $n=33$ ), as it can be used on an incomplete dataset to produce estimates which allow for the fit of a model to an entire sample (Little, Jorgensen, Lang & Moore, 2013).

The analysis was conducted in four stages. First, a Confirmatory Factor Analysis (CFA) was fitted to assess the construct validity of a latent variable of family functioning, using the 5 items of the Family APGAR scale. Model fit was assessed using a number of goodness of fit indexes including the Tucker Lewis Index (TLI), Comparative Fit Index (CFI), the Root Mean Square Error of Approximation (RMSEA), and Standardized Root Mean Square Residual (SRMR). Hu and Bentler (1999) recommend a CFI and TLI value  $>.95$ , an RMSEA  $<.06$  to  $.08$ , and an SRMR value  $>.08$  indicate good fit between the model and observed data (Hu & Bentler, 1999; Schreiber, Nora, Stage, Barlow, & King, 2006).

Next, six subsystem relationship observed indicators (that represented composite measurements of parental relationship satisfaction, parental disagreement, parent-child conflict, parent-child closeness, sibling warmth, and sibling conflict) were entered into the model and were covaried with the latent family functioning

construct and with one another. The strength of the correlations within the correlation matrix was used to determine which subsystem indicators would be entered into the structural models. Structural models were built to examine associations between the relationship indicators and family functioning. Subsystem indicators were entered sequentially into the model based on the strength of their association with family functioning in the correlation matrix. Finally, a within-sample replication was conducted to gauge factor stability.

## Results

### Measurement Model

The measurement model of the five Family APGAR items showed good fit to the data  $\chi^2(5) = 13.692, p = .018$ , CFI = .99, TLI = .98, RMSEA = .06 (90% CI .02 - .10). All factor loadings were significant ( $p < .05$ ) and ranged from .72 to .80 (Figure 4.1). Standardized factor loadings showed that Affection (APGAR4, Figure 4.1) had the strongest loading on family functioning ( $\beta = 0.80$ ).

Figure 4.1. *Confirmatory factor analysis of the latent construct of family functioning*

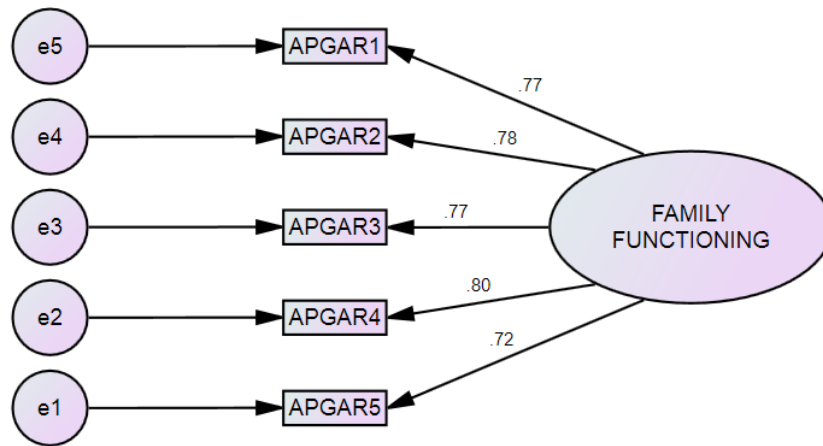


Figure 4.1. APGAR1, Adaption; APGAR2, Partnership; APGAR3, Growth; APGAR4, Affection, APGAR5, Resolve.

The correlation matrix showed that all of the six subsystem relationship indicators were significantly correlated with the latent construct of family functioning ( $r = -.13$  to  $.45$ ) (Table 4.2) and were therefore entered into the structural models.

Table 4.2 Correlation matrix of relationship variables

Variables	1	2	3	4	5	6	7
1. Family functioning	-						
2. Relsat	.45***	-					
3. Disagree	-.36***	-.40***	-				
4. CPCConflict	-.23***	-.07	.31***	-			
5. CPClose	.20***	.09	-.14**	-.28***	-		
6 .SRQConflict	-.13***	-.07	.31***	.43***	.10*	-	
7 .SRQWarmth	.24*	.10*	-.14**	-.25***	.40***	-.02	-

Note. *Relsat*, Global measure of relationship satisfaction; *Disagree*, How often couples disagree over issues related to their child; *CPCConflict*, Child-Parent Relationship Scale-Short Form Conflict Factor; *CPClose*, Child-Parent Relationship Scale-Short Form Closeness Factor; *SRQConflict*, Sibling Relationship Questionnaire Short version Conflict Factor; *SRQWarmth*, Sibling Relationship Questionnaire Short version Warmth Factor

\*  $p < .05$ , \*\*  $p < .01$ , \*\*\*  $p < .001$

## Structural Model

Each indicator was entered into the model sequentially, with paths drawn from each of the exogenous variables to the endogenous variable of family functioning. Models where all paths were significant were examined by a nested model comparison (Chi-Square difference test) to ascertain which one was most parsimonious (Preacher, 2006). As shown in Table 4.3, model 5 (including partner relationship satisfaction, partner disagreement, child-parent conflict, and sibling relationship warmth) accounted for the most variance in family functioning (29%). The chi-square comparison with model 3 was significant ( $p=.041$ ) indicating that model 5 including sibling warmth was more parsimonious than model 3. As the chi-square comparison demonstrates (Table 4.3), model 4 including child parent closeness (CPClose) was not as parsimonious as model 3 and so therefore CPClose was not included in model 5.

Standardised regression weights (Figure 4.2) showed that partner relationship satisfaction ( $\beta = .37, p < .001$ ) had the strongest positive association with family functioning, followed by sibling relationship warmth ( $\beta = .16, p < .001$ ). Disagreement between parents over issues related to the child ( $\beta = -.15, p = .004$ ), and conflict in the child-parent relationship ( $\beta = -.12, p = .016$ ) had a negative association with family functioning. The model showed good fit to the data  $\chi^2(21) = 36.544, p = .019$ , CFI = .98, TLI = .98, RMSEA = .04, and was accepted as the final structural model (Figure 4.2).

*Table 4.3 Structural models predicting family functioning*

Structural models	$\chi^2(df)$	CFI	TLI	RMSEA	SRMR	Total variance explained (R <sup>2</sup> )	All paths sig (p<.05)	Nested Chi-square comparison
1 – Relsat	15.526(9)	.99	.99	.04	.01	.20	yes	
2 – Relsat, Disagree	16.481(13)	.99	.99	.02	.01	.24	yes	
3 – Relsat, Disagree, CPConflict	26.628(17)	.99	.98	.03	.02	.26	yes	3 vs 2*
4 – Relsat, Disagree, CPConflict, CPClose	30.007(21)	.99	.98	.03	.02	.28	yes	4 vs 3
5 – Relsat, Disagree, CP Conflict, SRQWarmth	36.544(21)	.98	.98	.04	.02	.29	yes	5 vs 3*
6 – Relsat, Disagree, CPConflict, SRQWarmth, SRQConflict	46.181(25)	.98	.97	.04	.02	.29	no	

*Note.* \*p < .05

Figure 4.2. *Final structural model predicting family functioning*

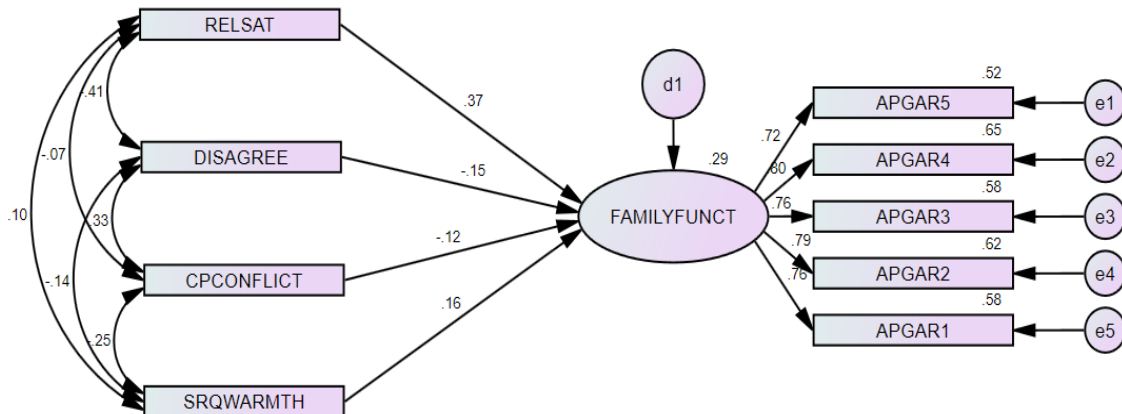


Figure 4.2. *Relsat*, Global measure of relationship satisfaction; *Disagree*, How often couples disagree over issues related to their child; *CPConflict*, Child-Parent Relationship Scale-Short Form Conflict Factor; *SRQWarmth*, Sibling Relationship Questionnaire Short version Warmth Factor

As recommended by Pohlmann (2004), a within-sample replication was performed to gauge factor stability. We randomly divided the sample into two groups (Group 1  $n=215$ , Group 2  $n=216$ ) and fitted the final structural model to both halves of the dataset. Model fit for Group 1 ( $\chi^2(21)=34.843$ ,  $p=.029$ , CFI = .98, TLI = .96, RMSEA = .05) was good (Table 4.4). Model fit for Group 2 was alarmingly good ( $\chi^2(21)=17.764$ ,  $p=.664$ , CFI = 1.00, TLI = 1.01, RMSEA = .00) (Table 4.4), which may be a result of low statistical power failing to detect discrepancies between the observed and specified model. However, within the models the directions of the paths in both the random groups were the same as within the overall sample (Table 4.5).

*Table 4.4 Within-sample structural models predicting family functioning*

Structural models	$\chi^2(df)$	CFI	TLI	RMSEA	SRMR	Total variance explained ( $R^2$ )	All paths sig ( $p < .05$ )
1 – Overall sample	36.544(21)	.98	.98	.04	.02	.29	yes
2 – Random Group 1	34.843(21)	.98	.96	.05	.03	.28	no
3 – Random Group 2	17.764(21)	1.00	1.01	.00	.02	.31	no

*Table 4.5 Within-sample associations between family functioning and relationship variables*

Structural models	Relsat $\beta$	Disagree $\beta$	CPConflict $\beta$	SRQWarmth $\beta$
1 – Overall sample	.37***	-.15**	-.12*	.16***
2 – Random Group 1 ( $N=215$ )	.30***	-.14	-.16*	.21**
3 – Random Group 2 ( $N=216$ )	.43***	-.17*	-.06	.10

*Note.* \*  $p < .05$ , \*\*  $p < .01$ , \*\*\*  $p < .001$



## Discussion

The current study explored whether dimensions of dyadic relationships in the family were related to mothers' perceived family functioning. The factor structure of family functioning among families of children with ID was initially tested in a CFA where the five observed Family APGAR variables (Adaption, Partnership, Growth, Affection, and Resolve) loaded onto a latent construct of family functioning. Good model fit suggested that family functioning is a valid construct that can be described using the Family APGAR items in this sample of mothers of children with ID. Given that the Family APGAR is a relatively new measure to be used in the ID field, further validation work is needed to fully establish its psychometric properties. However, the data suggest that it might be a useful measure for research which explores family functioning in families of children with ID.

In terms of our main research aim, the final SEM suggested that four family subsystem dimensions representing the three family subsystems studied: partner relationship satisfaction, sibling warmth, partner disagreement, and child-parent conflict were all associated with family functioning, accounting for 29% of the variance in the family functioning latent construct. Our findings support general predictions derived from FST and specifically the suggestion of 'spillover' (Erel & Burman, 1995) in that at least one relationship variable within the three subsystems had a significant association with the broader construct of family functioning. Despite much theoretical discussion about the family consisting of interconnecting subsystems, very little empirical research has explored whether these subsystems are related to a broader-level construct of family.

Our findings show that both of the variables capturing aspects of the marital/partner relationship were predictive of family functioning, with the quality of the relationship between the mother and their spouse or partner the strongest predictor of family functioning. This supports previous work which has reported a relationship between family adjustment/cohesion and marital adjustment/satisfaction in families of children with ID (Trute, 1990), and marital satisfaction and overall family functioning of mothers in the general population (Kitzmann, 2000). The

finding also strengthens FST which postulates that the couple subsystem is at the heart of the family unit (Seligman & Darling, 2007).

Our findings also show a positive relationship between the sibling subsystem and the broader family subsystem, with perceived warmth between the child with ID and their sibling a significant predictor of how mothers report on family functioning. This is an important finding given the lack of empirical work exploring the relationship between the sibling subsystem and the wider family system, and suggests that the presence of a positive, warm sibling relationship has some bearing on how mothers perceive their family to be functioning.

The finding that conflict in the parent-child relationship is negatively associated with how mothers report on the functioning of the family may reflect previous evidence of an association between the increased behavioural and emotional problems in children with ID and poorer family functioning (Herring et al., 2006; Jellett et al., 2015). While the study did not focus on behavioural and emotional problems per se, one might theorise that there is a spurious association whereby child behavioural and emotional problems increase parent-child conflict which then influences how mothers feel about the functioning of their family. More conflictual relationships are likely to have an impact on certain aspects of family functioning captured in the Family APGAR, such as the way that family members express affection and respond to emotions, and the way that the family share time together.

This study has provided an initial insight into the associations between dimensions of subsystem functioning and maternal perceptions of family functioning. As the data were cross-sectional, future replication is needed, with longitudinal data, to allow exploration of causal pathways. At this point, it is unclear why certain relationship dimensions were more strongly associated with family functioning than others. It is also important to note a number of other study limitations, namely that our findings only capture perceptions of family functioning and not necessarily how the family is actually functioning. Diary studies may be a way of overcoming this limitation which have been used in research exploring the influences of maternal perceptions of the child and marital adjustment on wellbeing

in mothers of children with autism (Lickenbrock, Ekas, & Whitman, 2011). The diagnostic status of the child with ID and siblings included in this study were based on parent report which could affect validity and specificity of the findings, however, we are confident that the majority of the study sample were recruited via a charity for children with neurological conditions. It is also acknowledged that this study focuses on the experiences of mothers who are currently in a partner relationship, and with at least two children, and that therefore the findings cannot be extended to single mothers, mothers with one child, or fathers. Diverse family structures and subsystems will have an effect on family functioning outcomes. Further work should look to understand family functioning in other family structures. Finally, there is also a problem of source variance in that mothers provided data on family functioning and their perceptions of the putatively contributing family subsystems.

The current study was designed to illustrate the potential for research to inform the associations between family subsystem and broader family level functioning in mothers of children with ID. It showed that mothers' reports of partner relationship satisfaction, sibling warmth, partner disagreement, and ID child-parent conflict were all associated with their perceptions of family functioning. Such research has the potential to enhance our theoretical understanding and to develop an evidence-base to inform family systems-based interventions. Specifically, interventions which target the marital/partner subsystem could improve the functioning of the family unit.

**Chapter 5: Psychological Wellbeing in Single Mothers of Children with  
Intellectual Disability (ID)**

### Abstract

Despite evidence which indicates that single mothers are more likely to be parents to a child with a disability, ID research has predominantly reported on the outcomes of mothers who live in two-parent households. The present study explored the psychological wellbeing of 213 single mothers of children with ID aged 4-15 years, using primary data from the Cerebra 1000 Families Study, a large-scale survey of parental caregivers of children with ID in the UK. Drawing on evidence from general population and two-parent family ID research, the study investigated whether a range of mother, child, and family-level factors were related to maternal psychological distress (depression and anxiety) and maternal life satisfaction. Regression analyses revealed that socioeconomic position (SEP) was negatively associated with mothers' reports of psychological distress and positively associated with life satisfaction. Impact of caring on mothers' personal lives and child behavioural and emotional problems were also found to be positively associated with psychological distress. Positive gain related to parenting the child with ID, and having a child with additional needs, were positively associated with mothers' life satisfaction. These findings demonstrate the impact that factors unrelated to the child with ID, namely socioeconomic position, can have on single mothers' psychological distress and life satisfaction, corroborating work with mothers in two-parent families. The findings also suggest that we should continue to support for factors related to the child, such as challenging behaviour and impact of caring on mothers' personal life, and also consider how we can bolster positive factors, such as positive gain. Given that this is the first study to explore correlates of psychological wellbeing in single mothers of children with ID, further work is needed to better understand their outcomes and support needs.

**Key words:** Single mothers, intellectual disability, autism, wellbeing

## **Introduction**

The psychological wellbeing of mothers has been the focus of a wealth of research in the field of Intellectual Disability (ID). Research findings suggest that raising a child with ID can have a significant negative impact upon the wellbeing of mothers and their family (Hastings, 2016). However, the majority of this research has focused on mothers of children with ID who live within two-parent households (Levine, 2009). A paucity of research which pays attention to the experiences of single mothers in this population is incongruent with evidence that suggests that single mothers are more likely to be parents to a child with a disability (Fujura & Yamaki, 2000). A study of parental separation, partnering and re-partnering among a UK population-based cohort found that children at risk of intellectual or developmental delays were significantly less likely than other children to be living in households with both biological parents, or in households where the mother was married when the child was aged 9 months, 3 years and 5 years old (Hatton et al., 2010).

Family Systems Theory (FST) is a theoretical framework that has often been used to help us understand wellbeing in family systems (Cox & Paley, 1997; Seligman & Darling, 2007). However, FST has often been interpreted or framed in the context of two-parent families, and the marital/partner subsystem has been proposed as the heart of family unit (Seligman & Darling, 2007). Thus, FST has been applied in a manner that potentially fails to recognise diverse family types and structures.

The relationship between single-parent status and psychological wellbeing is well documented, with research showing increased levels of mental health problems among single mothers compared to partnered mothers (Butterworth, 2004; Crosier, Butterworth & Rodgers, 2007; Targosz et al., 2003; Wang, 2004). Single mothers have also been found to report poorer health outcomes than partnered mothers (Rousou, Kouta, Middleton, & Karanikola, 2013) and lower levels of life satisfaction compared to partnered mothers and childless singles (Pollmann-Schult, 2018). There has been some work which has included single mothers of children with ID either through comparative study (versus partnered mothers of children with ID) or through

the control of single-parent status in data analyses. In line with findings in the general population, single mothers of children with ID have been found to report poorer psychological outcomes when compared to partnered mothers of children with ID (Blacher, Lopez, Shapiro, Fusco, 1997; McConkey, Truesdale-Kennedy, Chang, Jarrah & Shukri, 2008; Olsson & Hwang, 2001; 2008).

Moving beyond demonstrating significant between-group differences, it is important to begin to understand determinants of wellbeing in single parents. However, studies on wellbeing in these families that simply control for single-parent status do not give the breadth or power to investigate the outcomes for single parents specifically. Other than structural family differences, single parenthood may be associated with other factors that are important for parents' psychological wellbeing. However, no previous research reporting on the correlates of psychological wellbeing in single mothers of children with ID could be found. To inform the factors to be explored in the current research, a brief overview of key variables in research with single mothers in the general population and other research on the psychological wellbeing of parents of children with ID, will be discussed. The term psychological wellbeing used here refers not only to symptoms of psychological distress (depression, anxiety) but more broadly to include other dimensions of wellbeing such as life satisfaction. As stated by the World Health Organization, mental health and wellbeing is a "state of complete physical, mental and social-wellbeing and not merely the absence of disease or infirmity" (WHO, 2014, para.2).

One possible correlate of psychological wellbeing for single mothers of children with ID is socioeconomic status (SES). Differences in psychological wellbeing and health between single and partnered mothers of children without ID has been consistently linked to measures of socioeconomic status (Targosz et al., 2003) and hardship (Hope, Power, & Rodgers, 1999; Rousou et al., 2013). Crosier et al. (2007) found that 94% of the association between single mother status and poor mental health was accounted for by factors related to socioeconomic status and social support. Families of children with disabilities are more likely to experience socioeconomic disadvantage compared to families without a disabled child (Emerson, 2003; Parish & Cloud, 2006). An international meta-analysis by Spencer, Blackburn, and Read (2015) concluded that children with ID were at increased risk

of living in a low SES household. Families of children with ID are also at greater risk of persistent poverty. Emerson (2004) describes the “downward social mobility” (p.324) of families who raise a child with ID, shown to be associated with the financial and social impact of caregiving (Emerson, 2007). There is also consistent evidence that socioeconomic disadvantage is a significant predictor of maternal wellbeing in families of children with ID (Emerson, 2003; Emerson, 2004; Emerson et al., 2010; Olsson & Hwang, 2008). Single mothers of children with ID will be both primary caregiver and wage earner in their household (Taylor & Conger, 2017), therefore it is important that to explore socioeconomic position (SEP) as a correlate of their wellbeing.

Other factors of interest relate to characteristics associated with the child with ID. Children with ID are at heightened risk of behaviour problems (Eisenhower et al., 2005; Emerson & Hatton, 2007), which in existing research has been consistently found to pose a risk for the psychological wellbeing of parents (Hassell, Rose & McDonald, 2005; Hastings, 2002; Herring et al., 2006; Kersh et al., 2006; Sloper, Knussen, Turner, & Cunningham, 1991). A diagnosis of autism in addition to ID in the child has also been shown to be associated with elevated psychological problems for mothers (Griffith, Hastings, Nash, & Hill, 2010; Totsika et al., 2011a, 2011b). The additional needs of the child may also be associated with single mothers’ wellbeing. ID can coexist with a number of other conditions, such as epilepsy, physical health problems, and sensory impairment (Carvill, 2001). However, studies have neglected to explore the cumulative impact of a child’s additional needs as a predictor of wellbeing in single mothers of children with ID. It could be hypothesised that mothers of children with the greatest level of additional need may be at risk of poorer outcomes.

It is also important that variables directly associated with caring for a child with a disability are also considered. Informal carers of individuals with ID have been found to have a higher caring load compared to other carers and report a small negative impact on personal life (relationships with other people, spare time and hobbies) comparable to that reported by carers of people with mental health problems or dementia (Totsika et al, 2016). Single mothers are more likely to have increased caring responsibilities and be less able to engage in social and leisure



activities due to the absence of support from another caregiver. A mother's perceived impact of caring on her personal life may be associated with her wellbeing: if she is unable to engage in activities outside of her caring role due to caring demands then this may have negative implications for psychological distress and how satisfied she is with her life overall.

Conversely, it is also important to explore whether positive aspects related to parenting a child with ID are correlated with mothers' wellbeing. Parents of children with ID also report positive experiences associated with raising their child (Hastings & Taunt, 2002) and positive gain associated with parenting a child have been studied in the ID field (Griffith et al., 2010; Jones et al., 2013; MacDonald et al., 2010). However, currently there are no research studies where a positive gain variable has been used to predict the psychological distress and life satisfaction outcomes of single mothers of children with ID.

The aim of the present study was to describe correlates of psychological wellbeing in single mothers of children with ID. Drawing on evidence from the general population and two-parent family ID research, a range of mother, child, and family-level factors were investigated. The study explored whether these correlates were related to maternal psychological distress (depression, anxiety) but also to maternal life satisfaction.

## Method

### Participants

Two-hundred and thirteen single mothers of a child with ID participated in this study (see Table 5.1). The majority of mothers ( $n=194$ , 91.1%) were biological parents of the child with ID. One hundred and twenty-two mothers (57.3%) were educated below university degree level, and one-hundred and twenty-six (59.2%) were not in paid employment at the time of the research. One-hundred and nineteen mothers (55.9%) reported that they did not have a longstanding illness, disability or infirmity. The majority of mothers described themselves as White British: 194 (91.1%). The modal weekly household income was £201-400: 117 (54.9%). Mothers had on average one child living in the household ( $M=1.84$ ,  $SD=0.95$ , Range =1-5). Most mothers said they were managing financially: 173 (81.2%), however the majority of mothers could not raise £2000 in an emergency: 112 (52.6%). A socioeconomic position (SEP) composite showed that 15 (7.0%) mothers were categorised into the lowest scoring group of zero (not educated to university level, not in employment, financially struggling, and could not raise £2000 in an emergency). Fifty-six (26.3%) mothers comprised the second lowest group, scoring one indicator of higher SEP; sixty-one (28.6%) mothers had two high SEP indicators; fifty mothers (23.5%) had three high SEP indicators, and thirty-one (14.6%) were in the highest SEP group with four indicators.

Mothers reported on the severity of their child's ID. One-hundred and two (47.9%) children were reported to have 'mild/moderate' ID, and 108 (50.7%) 'severe/profound' ID. A bit over half of the children also had ASD ( $n=119$ , 55.9%). Children were on average 9 years-old ( $SD=2.93$ , Range=4-15 years) and the majority were male: 153 (71.8%). Children were reported to have a range of additional needs including visual ( $n=57$ , 26.8%) and hearing ( $n=27$ , 12.7%) impairments, epilepsy ( $n=35$ , 16.4%), mobility ( $n=136$ , 63.8%), and physical health problems ( $n=115$ , 54.0%). An additional needs index was created with scores ranging from 0-5. Most children had two additional needs: 63 (29.6%).

*Table 5.1 Demographic profile of participating single mothers (N=213) and their child with intellectual disability*

Variables	%
Mother status	
Biological	91.1
Step	7.0
Foster	1.9
Education level	
University degree level or above	42.7
Below degree level	57.3
Employment status	
In work	40.8
Not in work	59.2
Ethnic group	
White	91.1
Mixed	1.4
Indian	0.5
Pakistani and Bangladeshi	0.5
Black or Black British	2.8
Other Ethnicity	2.8
Missing	0.9
Mother illness/disability/Infirmary	
Yes	44.1
No	55.9
Weekly household income	
£200 or less	5.2
£201-400	54.9
£401-600	24.4
£601-800	8.9
£801-1000	1.4
£1000+	2.3

Variables	%
Subjective poverty	
Managing	81.2
Struggling	18.8
Hardship/could raise £2000	
Yes	47.4
No	52.6
Child ID status	
Mild/Moderate	47.9
Severe/profound	50.7
Missing	1.4
Child disability	
Autism	55.9
Child gender	
Male	71.8
Female	27.7
Missing	0.5
	Mean (SD)
Age of child with ID	9.54 (2.93)
Number of all children in the household	1.84 (0.95)
Number of all people in the household	3.07 (1.05)

Note. SD: Standard Deviation

## Maternal Measures

The Kessler 6 (K6) (Kessler et al., 2003) is a 6-item measure of psychological distress (see Appendix S, p.279). It is a well-validated measure which has been used in US National Health and Household surveys (Kessler et al., 2010) and is an established measure within studies of parents of children with ID (Emerson et al., 2010) and ASD (Weiss & Lunskey, 2011). The K6 asks respondents to rate how frequently they have experienced symptoms of psychological distress (i.e. feeling “so depressed that nothing could cheer you up?”) during the last 30 days on a Five-point Likert scale from *none of the time (0)* to *all of the time (4)*. Items are summed

to give a total score ranging from 0-24, with higher scores indicating increased levels of psychological distress. In addition, a cut-off score of 13 and above is indicative of serious mental illness (SMI) using DSM-IV criteria (Kessler et al., 2003). Internal consistency in the present sample was very good (Cronbach's  $\alpha$ : .87).

Life satisfaction was measured using a single item which asked mothers to rate "how satisfied they were with the way their life had turned out so far" on a scale of 0 (*extremely unsatisfied*) to 10 (*completely satisfied*). This measure has been used in large UK and European population studies to measure subjective wellbeing across populations, and has been analysed in relation to mothers of children with ID (Totsika et al., 2011a). Higher scores indicate greater satisfaction with life, and scores can be categorised as low (0-4), medium (5-6), high (7-8), and very high (9-10) (ONS, 2017).

The impact of caring for a child with ID on a mother's personal life (termed here as impact on personal life) was assessed by asking mothers whether their ability to spend time doing specific leisure and social activities had been affected by the assistance that they give to their child with ID (see Appendix T, p. 280). Mothers were presented with seven options including "Reduced time with friends" and "Unable to socialise or take part in social or leisure activities at all" and asked to select any that applied. An index of impact on personal life was created by counting items where mothers indicated impact was present. Scores on impact on personal life ranged zero to seven with higher scores indicating greater impact on personal life. This measure has been used in a 2009-10 survey of carers in households in England (NHS Information Centre for Health and Social Care, 2010) and in research with informal carers of people with ID (Totsika et al., 2016).

The Positive Gain Scale (PGS; Pit-ten Cate, 2003) was used to explore mothers' perceived benefit of raising a child with ID (see Appendix U, p.281). The PGS requires respondents to rate seven items on a Five-point Likert scale ranging from *strongly agree* (1) to *strongly disagree* (5). Items include: "Since having this child I have grown as a person" and "Raising this child has put my life into perspective". Scores are reversed to give a total score (range 5 to 35), with higher scores indicating more perceptions of positive gain. The PGS was developed

following interviews with parents with disabilities (Pit-ten Cate, 2003) and has been used in a number of studies with this population (Jones et al., 2013; Macdonald et al., 2010; Weiss & Lunsky, 2011). Internal consistency in the present study was good (Cronbach's  $\alpha$ : .79).

A socioeconomic position (SEP) variable was created based on four measures: maternal education, maternal employment, subjective poverty, and hardship. The subjective poverty measure required mothers to rate how well they were managing financially i.e. "How well would you say you are managing financially these days?" on a Five-point Likert scale from *living comfortably (1)* to *finding it very difficult (5)*. The measure of hardship required mothers to state how easy it would be for them to raise £2000 in an emergency, i.e. "Suppose you only had one week to raise £2000 for an emergency, which of the following best describes how hard it would be for you to get that money?" on a Four-Point Likert scale from *I could easily raise the money (1)* to *I don't think I could raise the money (4)*. All measures were dichotomised into two groups (0/1). Mother education: 0=educated below degree level, 1=educated to degree level or higher. Mother employment: 0=not in employment at the time of the research, 1=in employment at the time of the research. Subjective poverty: 0=financially struggling (scores 4 and 5), 1=financially managing (scores 1 to 3). Hardship: 0=could not raise the money (scores of 4), 1=could raise £2000 (scores of 1 to 3). Scores for each of the variables were then summed (0-4) to give a socioeconomic position of 0 (low SEP: scoring 0 on all four of the indicators) to 5 (high SEP: scoring 1 on all four of the indicators).

## **Child Measures**

The Strengths and Difficulties Questionnaire (SDQ; Goodman, 1997) is a measure of child behavioural and emotional problems (see Appendix R, p.278). The 25-item scale generates scores for emotional symptoms, conduct problems, hyperactivity, and peer problems and pro-social behaviours. A total behaviour problems score is obtained by summing the first four subscales. Caregivers indicate the extent to which each statement (e.g. "Often unhappy, downhearted or tearful") applies to the child on a Three-point scale: (*Not true, somewhat true, very true*) based on their child's behaviour over the past six months. The SDQ is a reliable

measure of child behavioural and emotional problems which has been used in research on children with ID (Totsika et al., 2011a, Totsika et al., 2011b). The total difficulties score was used in the present study, with a higher score indicative of greater behavioural and emotional problems. Internal consistency was good (Cronbach's  $\alpha$ : .70).

## **Procedure and Analysis**

The present analysis uses data from a large UK survey of parents of children with ID aged 4-5 years known as 'The 1,000 families study.' Following NHS ethical approval (NRES West Midlands, ref: 15/WM/0267) (see Appendix A, p. 150), study information was distributed via a number of third-sector organisations who support families of children with developmental disabilities. Recruitment took place primarily online via social media platforms and through local parent support groups. A primary caregiver was asked to complete a 20 minute online or paper survey, followed by a telephone interview. A total of 1082 caregivers took part in the study. The present research focuses on 213 mothers who reported that they were single, divorced, separated, widowed, or not currently living with a partner. As the aim was to capture mothers who were raising their children without a partner in the household, it was not possible to analyse the data according to the type of single-parent status.

Multi-variable linear regression models were conducted separately for psychological distress and life satisfaction to analyse the main effects of the following variables: impact on personal life, positive gain associated with parenting the child with ID, socioeconomic position, number of children in the household, mother illness/disability, child behavioural and emotional problems, child autism, and additional child needs.  $R^2$  was used to assess model fit. Analyses were conducted in SPSS Statistics 24.0®

## Results

### Descriptive Statistics

Table 5.2 presents the mean scores for mothers on measures of psychological distress (K6), life satisfaction, impact on personal life, and positive gain. It also presents the mean score for child behavioural and emotional problems (SDQ). The total psychological distress mean was 10.11 (SD: 5.61, Range 0-24), with 30.2% mothers scoring above the cut-off for SMI (13). Mean life satisfaction was 5.62 (SD: 2.14, Range 0-10) with the majority of scores falling into the very low (29.2%), medium (31.1%) and high life satisfaction categories (33.0%). The mean score for impact on personal life was 4.32 (SD=1.90, Range=1-7) and for the positive gain scale 28.09 (SD=4.33; Range=10-35). Mean SDQ total problem score was 18.85 (SD: 5.43, Range 0-35).

*Table 5.2 Mean scores on the Kessler-6 (K6), Life Satisfaction, Impact on Personal Life, Positive Gain Scale (PGS), and the Strengths and Difficulties Questionnaire (SDQ)*

Measure	Mean (SD)
K6	10.11(5.61)
Life satisfaction	5.62 (2.14)
Impact on personal life	4.32 (1.90)
PGS	28.09 (4.33)
SDQ	18.85 (5.43)

*Note. SD: Standard Deviation; K6, Kessler 6; PGS, Positive Gain Scale; SDQ, Strengths and Difficulties Questionnaire*

A correlation matrix of the predictor variables (Table 5.3) suggested some weak correlations between variables used to measure negative and positive impacts of parenting (impact on personal life, positive gain) and factors related to the child with ID (child SDQ, autism, additional needs), however examination of tolerance and variation inflation scores yielded no evidence of multicollinearity (Brace, Snelgar & Kemp, 2009).



*Table 5.3 Correlation matrix of Predictor Variables*

Variables	1	2	3	4	5	6	7	8
1. Impact	-							
2. Positive Gain	-.20**	-						
3. SEP	-.01	.03	-					
4. No. children	.06	-.11	.00	-				
5. Mother disability	.06	-.12	-.09	-.12	-			
6. Child SDQ	-.06	.03	-.25	.02	.11	-		
7. Child autism	.11	-.09	-.09	.05	.06	.32**	-	
8. Additional needs Index	.06	.04	-.08	-.12	.09	-.24**	-.33**	-

Note \*\* p < .01

## Regression Models

The K6 model was significant:  $F(8,190) = 6.085, p < .001$ , however it had low explanatory power, accounting for 20% of the variance in psychological distress ( $R^2 = .20$ ). Standardised betas showed small effects (Table 5.4). Impact on personal life was positively associated with mothers' psychological distress ( $\beta = .22, p = .001$ ), suggesting that as the impact on personal life increased, maternal psychological distress scores also increased. Socioeconomic position was negatively associated with psychological distress ( $\beta = -.19, p = .005$ ), suggesting that a lower socioeconomic position was associated with greater psychological distress. Child SDQ scores were positively associated with maternal psychological distress ( $\beta = .15, p = .034$ ), indicating increased psychological distress for mothers where their child had higher levels of child behavioural and emotional problems. Positive gain ( $\beta = -.07, n.s.$ ), number of children in the household ( $\beta = .05, n.s.$ ), mother illness/disability ( $\beta = .10, n.s.$ ), child autism ( $\beta = .04, n.s.$ ), and child additional needs ( $\beta = -.13, n.s.$ ), were not statistically significant.

A significant life satisfaction model emerged:  $F(8,190) = 3.659, p = .001$ , however again this had low explanatory power, accounting for 13% of the variance in life satisfaction ( $R^2 = .13$ ). Standardised betas showed small effects (Table 5.4). Positive gain was positively associated with mothers' life satisfaction ( $\beta = .20, p = .004$ ), suggesting that as the positive gain score increased, life satisfaction scores increased. Socioeconomic position was positively associated with life satisfaction ( $\beta = .16, p = .020$ ), indicating that a higher socioeconomic position was associated with greater life satisfaction. Child additional needs were positively associated with life satisfaction ( $\beta = .15, p = .037$ ), indicating greater life satisfaction where children have more additional needs. Impact on personal life ( $\beta = -.10, n.s.$ ), number of children in the household ( $\beta = -.07, n.s.$ ), mother illness/disability ( $\beta = -.03, n.s.$ ), child behavioural and emotional problems ( $\beta = -.00, n.s.$ ), and child autism ( $\beta = -.01, n.s.$ ) were not statistically significant.

Overall, factors related to parenting a child with ID had the strongest effect on mothers' outcomes: impact on personal life ( $\beta = .22$ ) had the strongest effect on mothers' psychological distress, and positive gain ( $\beta = .20$ ) had the strongest effect

on life satisfaction. SEP had similar strength effects on mothers' psychological distress ( $\beta = -.19$ ) and life satisfaction ( $\beta = .16$ ). Factors related to the child had weak positive associations with mothers' outcomes in both models, for example, child SDQ was associated with psychological distress ( $\beta = .15$ ), and child additional needs was associated with life satisfaction ( $\beta = .15$ ).

Table 5.4 Multiple regression analyses for variables predicting mothers' psychological distress and life satisfaction

	Psychological distress				Life satisfaction			
	<i>B</i>	<i>SE B</i>	$\beta$	<i>p</i>	<i>B</i>	<i>SE B</i>	$\beta$	<i>p</i>
<i>F</i>	6.085***				3.659***			
<i>R</i> <sup>2</sup>	.20				.13			
Impact of caring on personal life	0.67	0.20	.22	.001	-.11	0.08	-.10	.139
Positive Gain	-.10	0.08	-.07	.247	.10	0.03	.20	.004
SEP	-.92	0.32	-.19	.005	0.30	0.12	.16	.020
No. children	0.32	0.38	.05	.398	-.17	0.15	-.07	.260
Mother disability	1.13	0.75	.10	.133	-.13	0.29	-.03	.651
Child SDQ	0.16	0.07	.15	.034	-.00	0.03	-.00	.906
Child autism	0.51	0.82	.04	.531	-.08	0.32	-.01	.805
Child additional needs index	-.64	0.34	-.13	.061	0.28	0.13	.15	.037

Note. SEP, Socioeconomic Position; SDQ, Strengths and Difficulties Questionnaire

\*\*\*  $p < .001$

## Discussion

The current study is the first to explore correlates of psychological wellbeing in single mothers of children with ID. It is important that family research in this field recognises that not all children with ID are raised within two-parent families and the pertinence of exploring the wellbeing of single-parents of children with ID without the need for comparison.

Exploring factors that are associated with single mothers' psychological wellbeing enables us to understand their specific needs and experiences better. Our results show that socioeconomic position (SEP) was predictive of both mothers' psychological distress and life satisfaction, a finding which is consistent with previous studies which have reported the association between socioeconomic disadvantage and maternal wellbeing in mothers of children with ID (Emerson, 2003; Emerson, 2004; Emerson et al., 2010; Olsson & Hwang, 2008; Totsika et al., 2011a, 2011b). Families of children with disabilities are reported to experience greater socioeconomic disadvantage (Emerson, 2003; Parish & Cloud, 2006; Spencer et al., 2015) and being a single parent may further increase the likelihood of deprivation and associated wellbeing difficulties.

The results also showed that there were correlates of mothers' wellbeing which were related to parenting. The impact of caring for a child with ID on a mother's personal life was significantly associated with psychological distress. As previous work has found (Totsika et al., 2016), informal caregivers of children with ID report negative impacts on their personal life, and in this study mothers for whom caring had the largest impact on personal life, reported greater levels of psychological distress. Despite the demands of caring for a child with a disability being widely known, research has paid little attention to how these demands affect a mother's personal life and subsequent wellbeing. These findings highlight the importance of exploring the impact on mothers' personal life further due its implications for psychological distress.

The finding that child behaviour and emotional problems were found to be predictive of mothers' psychological distress has also been consistently reported in previous studies with parents of children with ID (Hassell et al., 2005; Hastings, 2002; Herring et al., 2006; Kersh et al., 2006) and further highlights the putative impact that challenging behaviour can have on parental wellbeing and the need for effective and timely intervention. It is interesting that a child's behaviour and emotional problems were not related to how satisfied mothers were with their life, indicating that this negative impact on parenting may have less of a bearing on more positively-orientated constructs such as life satisfaction. However, this is in contrast to some existing research which suggests that there is a negative association between life satisfaction and child behaviour problems in mothers of children with ID (Shivers, Leonczyk, & Dykens, 2016; Totsika et al., 2011a) and ASD (Totsika et al., 2011a). There have also been studies which have found a negative association between a construct of maternal positivity (including life satisfaction as one indicator) and child behaviour problems in mothers of children with ID (Jess, Hastings, & Totsika, 2017), and positive impact and child behaviour problems of mothers of children with and without DD (Blacher & Baker, 2007). The finding that having a child with autism in addition to ID was not predictive of either maternal psychological distress or life satisfaction is in contrast to previous work (Griffith et al., 2010; Totsika et al., 2011a, 2011b), reinforcing the importance of orientating support towards behaviour problems as well as disability status.

The findings also show that positive aspects of parenting a child with ID are associated with how satisfied mothers are with their life, with positive gain one of the strongest predictors of maternal life satisfaction. It could be that mothers who are more satisfied with life tend to report more positive gain from raising a child with ID because these two constructs are related to a higher-level construct related to overall positivity: findings from a study of 135 mothers of children with ID which conducted a confirmatory factor analysis (CFA) found that an underlying positivity construct could

be described using five indicators of positivity, including mothers' satisfaction with life ( $\beta = 0.58$ ) and mothers' perceptions of their child's positive contributions to themselves and their wider family ( $\beta = 0.48$ ) (Jess et al., 2017). Another reason for the association could be that these mothers start with a higher level of life satisfaction prior to becoming a parent to the child with ID, and may then be more likely to report positively in regard to their parenting role. Future longitudinal studies are needed to capture these factors at multiple time points and understand the relationship between the two.

The positive association between a child's additional needs and the life satisfaction of single mothers is an interesting finding. While a negative association between these two variables may be expected, it perhaps reflects that mothers of children with the most additional needs may be receiving greater support. The study also did not explore whether mothers not in paid employment had children with more additional needs and so may therefore not have the double burden of work and care, however no group differences were found to support this hypothesis. There is no doubt that the needs of a child in addition to ID require far more exploration in relation to mothers' wellbeing. It would also be pertinent to explore whether this association is present for other members of the family such as fathers, in order to improve source variance and understand factors which are associated with their life satisfaction.

The study has provided an insight into some of the factors associated with the psychological wellbeing of single mothers of children with ID, however it is important to highlight some limitations. Firstly, single mothers of children with ID are a heterogeneous group and findings may vary depending on whether the single-parent status results from never-marrying, divorce, separation, the death of a partner, or not living with a partner. The survey aimed to capture mothers who were raising their children without a partner in the household whatever the reason of single parent status, and so it was not possible to analyse the data according to the type of single parent. In line with this, the survey did not capture detail on how long mothers had been a single

parent, whether the father of the child had a role in parenting, and what other formal or informal sources of support the mother may be receiving. Further research on the wellbeing of single parents of children with ID should look to capture and control for such variables.

The current study was designed to focus specifically on the psychological wellbeing of single mothers of children with ID, to identify correlates of their psychological distress and life satisfaction. While the outcomes of mothers with ID has received much empirical attention, research which has specifically explored the outcomes of single mothers in the field of family disability research has been extremely limited. This study has studied single mothers of children with ID as a family system in their own right, encouraging family researchers to recognise diverse family types and structures and develop systems frameworks which are specific to single-parent families.

Overall the findings suggest that correlates of wellbeing in single mothers were similar to mothers in two-parent families. Mothers' wellbeing in this study was related to factors which are unrelated to parenting a child with ID such as socioeconomic position and so support should continue to target single-parents most at risk of disadvantage. It has also identified that there are factors related to parenting (impact on personal life, positive gain) and the child (behaviour problems, additional needs) which can have positive and negative implications for single mothers' wellbeing. Professionals should continue to offer support for factors which have a negative impact on mothers' wellbeing, namely the impact of challenging behaviour, or of caring on a mother's personal life, yet also consider how they can bolster positive factors, such as positive gain. Future research should continue to explore the outcomes of single mothers of children with ID, including a broader range of factors to form a deeper understanding of their wellbeing, in order to adjust narratives which have implications for service delivery.



## **Chapter 6: General Discussion**

Family members of children with ID, like in any other family, have an influence on, and are influenced by, one another. This thesis has developed a more holistic understanding of families of children with ID by exploring parts and members of the family system where there is little existing knowledge. In the Introduction (Chapter 1), an overview of Family Systems Theory and existing literature in the field was provided, and methodological considerations when conducting family research were discussed. Following this, four empirical papers were presented (Chapters 2,3,4,5) which explored different aspects and members of the family system in families raising a child with ID which have received little empirical attention. This discussion will now provide a summary of the findings, outline implications of this thesis in relation to theory and practice in the field, and discuss limitations and future research directions.

### **Summary of Thesis Findings**

The introduction presented in chapter 1 discussed the need for a systemic approach to ID research. It argued that there has been a substantial body of work in the field on maternal outcomes and mother-child relationships, however research which explores the outcomes of other family members and the interactions between different subsystems within the family unit is still in its infancy. An appraisal of Family Systems Theory demonstrated that this is a useful conceptual framework which can be applied to families of children with ID. The review of systems-informed research in the ID field presented studies which have explored outcomes at the individual, subsystem, and family-unit level and concluded overall that there are many unexplored avenues in each of these levels, which this thesis aimed to address. The review concluded by outlining methodological considerations for future research, specifically, the need for: large sample sizes to be able to conduct analyses which examine family subsystems and include multiple family members; psychometrically robust measures and standardised ways of scoring; and statistical techniques which account for the interdependence of family members when conducting within-family analyses.

Chapter 2, the first empirical study, contributed new knowledge in regard to the relationship satisfaction of parents of children with ASD. The research utilised advanced multilevel modeling (MLM) techniques accounting for the interdependence of data provided by mothers and fathers in the same family, to explore whether mothers and fathers report similar or different levels of relationship satisfaction. The study then explored whether parental relationship satisfaction was associated with factors such as parental depression and the behavioural and emotional problems of a child with ASD and/or a sibling. The analysis showed that mothers and fathers of children with ASD reported similar levels of relationship satisfaction, and that relationship satisfaction was negatively associated with parental depression and child behavioural and emotional problems, even when controlling for other family factors such as socioeconomic status and number of children in the family. Overall mother-father couples were more likely to report relationship satisfaction in the ‘adjusted’ range. The majority of couples scored in the ‘normal’ category for depressive symptoms, however more mothers than fathers scored above clinical cut-off, and there were statistically significant gender differences in depression scores with mothers reporting higher depression scores than fathers. The study highlighted the importance of exploring the impact of raising a child with ID on subsystems that do not just include children, as increased parenting demands can have implications for parents in their role as a spouse or partner. The study also demonstrated the advantages of using sophisticated statistical techniques such as MLM when analysing nested family data.

Chapter 3 added to the limited literature exploring the psychological wellbeing of fathers with and without a child with ID. The study drew upon a large UK representative sample of fathers of children aged 5 years and explored whether fathers with and without a child with ID differed on measures of wellbeing (life satisfaction, work-family balance, general health) and parenting (competency, parent-child closeness). It subsequently examined whether the presence of a child with ID was still a significant predictor of paternal wellbeing when controlling for a number of correlates of wellbeing. While initial comparisons showed that fathers of children with ID reported

lower levels of life satisfaction and poorer general health compared to fathers of children without ID, having a child with ID was not a significant predictor of paternal wellbeing when controlling for a range of other variables. Overall, child behavioural and emotional problems, and living in poverty, were two significant predictors of paternal wellbeing. A population representative sample such as this provided us with the opportunity to compare large numbers of fathers with and without a child with ID and explore correlates of wellbeing, in order to start to develop a better understanding of the father-child subsystem.

The research described in chapter 4 is unique in that it explored how mothers' perception of the functioning of three different subsystems - mother-father, parent-child, child-sibling - related to overall family functioning. To our knowledge, this is the first study in the field to have investigated functioning at the subsystem level and its associations with broader family functioning. This exploratory study showed that functioning in dyadic relationships were associated with broader constructs of family functioning. Partner relationship satisfaction, partner disagreement, child-parent conflict, and sibling relationship warmth accounted for the most variance in family functioning, with partner relationship satisfaction having the strongest positive association with family functioning. The results reinforced the importance of analysing data at the subsystem and family unit level. Such research has the potential to not only enhance our theoretical understanding, but also begin to inform and develop systems-based intervention for families of children with ID.

The research described in chapter 5 has also made a distinct contribution to the literature in the ID field by being one of very few studies to have explored the psychological wellbeing of single mothers of children with ID. The study examined mother, child, or family-level factors that could be related to two aspects of wellbeing: depression and anxiety, and life satisfaction. The findings showed that socioeconomic position was negatively associated with maternal psychological distress (depression and anxiety) and positively associated with life satisfaction. The impact of caring for a child with ID on mothers' personal life, and child behaviour and emotional problems, were

positively associated with maternal psychological distress. Positive gain from parenting the child with ID, and the additional needs of the child with ID, were both found to be positively associated with life satisfaction. The study helps us to understand whether the psychological wellbeing of single mothers of children with ID is related to factors associated with wellbeing of mothers in the general population and in two-parent families of children with ID. Given that children with ID are more likely to live in single-parent households, the study reinforces the need to identify avenues of support for single mothers of children with ID.

### **Theoretical Implications**

The results that have emerged from this thesis confirm the overarching principle of Family Systems Theory that individuals and subsystems within the family are interconnected and have an influence on one another. The research described in Chapter 2 has been able to demonstrate the interdependence of family members of children with ID, with mothers and fathers in the same family reporting similarly on a measure of relationship satisfaction, supporting the theory of ‘crossover’ whereby one partner’s level of satisfaction transfers to the other partner in the household (Gernstein et al., 2009). The negative association between child behaviour problems and relationship satisfaction supports previous work (Benson & Kersh, 2011; Hartley et al., 2012; Sikora et al., 2013; Robinson & Neece, 2015) and indicates that maintaining boundaries between the marital/partner subsystem and parental subsystem may be a challenge when a child has challenging behaviour. Consistent with previous research with parents of children with ASD (Benson & Kersh, 2011; Shtayermman, 2013; Sim et al., 2016; Weitlauf et al, 2014), the results in Chapter 2 showed that mental health difficulties at the individual parent level, can impact outcomes, such as relationship satisfaction, at the subsystem level.

The results described in Chapter 4 have provided new evidence that subsystem functioning is associated with functioning at the broader family level. Specifically, the finding that dimensions representing three family subsystems (parent-parent, child-parent, child-sibling) were associated with the construct of family functioning in mothers of children with ID indicates that mothers' perceptions of functioning in one subsystem can effectively 'spill over' (Erel & Burman, 1995) into how she perceives the family to be functioning as a whole. The results of the study in Chapter 4, which also found that partner relationship satisfaction had the strongest positive association with family functioning, appears for these mothers to confirm the importance of the marital/partner subsystem for the overall functioning of their family. If such a finding was replicated in future work with fathers and/or other mothers one could hypothesise that there is some hierarchy within the family system, as the couple subsystem appears to take up a larger part of the construct of family when evaluating the wellbeing of the whole system in two-parent families. Despite the potential for disequilibrium in the family system when there is a child with ID, the evidence seems to suggest that the overall wellbeing of a family with a child with ID is associated more with the marital/partner subsystem than the parental one. It would be useful to explore whether the same conclusions are reached if the study was conducted with fathers. Evidence from Chapter 2 which demonstrated crossover between mothers and fathers in the same family on a measure of relationship satisfaction, suggests that this may be the case. It would also be interesting to understand how single parent families report on overall family wellbeing if the most prominent subsystem, the marital/partner subsystem, is absent.

Taken together, the findings from Chapters 3 and 5 which showed that elevated child behavioural and emotional problems were significantly associated with paternal wellbeing in fathers of children with and without ID (Chapter 3), and maternal psychological distress in single mothers of children with ID (Chapter 5), corroborate a plethora of work which has reported a negative association between child behavioural and emotional problems and maternal wellbeing in mothers of children with ID in two-

parent families. These chapters have also further emphasised the pervasive impact that socioeconomic status can have on parental wellbeing outcomes. The results from Chapter 3 showed that fathers living in poverty reported lower levels of psychological wellbeing, including lower life satisfaction and work-family balance, but also a more negative parent-child relationship. Chapter 5 demonstrated that there is an association between socioeconomic position and psychological wellbeing for single mothers of children with ID, with mothers of lower SEP reporting higher levels of distress (depression and anxiety) and lower levels of life satisfaction. These findings can be understood within the context of FST. ‘Variety’, the extent to which a “system has the resources to meet new environmental demands” (White et al., 2015, p.150) is proposed as necessary for families to adapt to challenges and ensure system equilibrium. However, if families do not have much variety, i.e. access to formal or informal support for their child’s behaviour problems or available economic resource, then these findings suggest that this could have implications for parental wellbeing.

The findings have also furthered understanding in regards to mother-father differences in families of children with ID. Like much previous research (Jones et al., 2013; Lee, 2009), the findings in Chapter 2 have also confirmed differences in mental health outcomes for mothers and fathers of children with ASD, with mothers reporting statistically higher depression scores compared to fathers. Chapter 3 has also started to begin to understand the wellbeing of underrepresented family members in the literature, such as fathers. It was interesting to find that father wellbeing was not related to parenting a child with ID and subsequently begin to reflect as to why this is in contrast to mothers where there is an established pattern of difference between mothers with and without a child with ID.

Chapter 5 also provides new knowledge in regard to other family structures, such as single mother households. FST frameworks focus on a family unit with two parents, however it is clear that there is a need to develop systems frameworks within the context of single-parent families. Furthermore, what we know currently about the outcomes of parents in the ID field is based upon evidence with mothers in two-parent families, and

so the findings from this chapter allow us to learn more about the specific wellbeing of mothers in single-parent households. The findings from Chapter 5 indicated that the factors affecting the wellbeing of single mothers were not distinctive, and instead were similar to those reported in studies with mothers in two-parent families and fathers, such as socioeconomic position and child behaviour problems. However, as the greatest predictors of single mothers' wellbeing were related to parenting their child with ID, we can hypothesise that single mothers' wellbeing may be more likely to be associated with aspects of the parent-child relationship. As the biggest predictor of mothers' psychological distress was the impact of caring for their child with ID on their personal life, it could be that the demands of caregiving are more acutely felt by single mothers who are likely to have greater caring responsibilities, and are thus more restricted in their ability to partake in leisure and social activities, due to difficulties accessing childcare. However, the findings also show that the association between parenting a child with ID and the wellbeing of single mothers is not all negative. As the perceived gain associated with raising a child with ID had the strongest positive association with mothers' life satisfaction, it suggests that these two constructs are in some way related and highlights the importance of exploring positive as well as negative aspects of parenting a child with ID.

Overall this thesis has rejected the prevailing narrative that raising a child with ID is always a negative experience. Most parents of children with ID did not score within clinically concerning ranges on measures such as psychological distress and relationship satisfaction, and overall scored well on measures of individual wellbeing such as life satisfaction and general health, and measures related to parenting such as parenting competency, parent-child closeness, and positive gain. While comparisons with normative data (in Chapters 2 and 3) typically showed that parents of children with ID fared worse compared to typical counterparts, this thesis highlights the importance of reviewing the outcomes of parents of children with ID in relation to clinical cut-off points. It was also clear from regression analyses that negative outcomes for parents



were not always directly related to the child with ID, highlighting the need to consider a broad range of factors.

### **Practical Implications**

Systemic approaches have obvious practical potential and are being increasingly applied when working with families of children with ID in the UK (Baum, 2007). At the broadest level, this thesis has shown that there is a continued need to support families of children with ID in a way which is family-centred and is inclusive of all family members and family types.

The combined findings from the empirical Chapters 2 to 5 have also demonstrated the need to challenge the simplistic and negative narratives which surround parenting a child with ID. These can be damaging for families and do not reflect the complex interplay of factors on many levels. Interventions which identify enablers and positive factors associated with parenting a child with ID, as well as endeavour to tackle the challenges and barriers, could have more progressive outcomes for families. It is also clear that it is important to adopt the broader World Health Organization definition of wellbeing when supporting families with children with ID, to examine aspects such as life satisfaction and not just the presence or absence of clinical depression or anxiety.

The findings from the four empirical chapters of this thesis have a number of practical implications. Firstly, there is a continued need to target child behavioural and emotional problems as this was a consistent predictor of individual parental and subsystem wellbeing. There was also emerging evidence from father analyses that we should be focusing more on child behaviour problems than a child's diagnostic label. This is something which could be ameliorated with the right support. There is also strong and consistent evidence for the need to support families of lower socioeconomic position, as most of the difficulties reported by parents were related to contextual factors

such as poverty. While this is certainly something which is more challenging to resolve at the individual level, actions applied at a broader policy level which seek to give those families with greatest need more options or ‘variety’ would go far in improving the lives of parents and in turn, the outcomes of their child with ID.

Analyses with two-parent families have demonstrated the importance of the marital/partner subsystem and therefore support in this subsystem could have consequent benefits for the rest of the family. For example, in Chapter 4 it was evident that partner relationship satisfaction had the strongest association with mothers’ perceptions of family functioning, therefore strengthening the marital/partner subsystem could benefit the wider family unit. In Chapter 2 it was also evident that there is potential for ‘spillover’ between parental and marital subsystems, highlighting the possible need for support which helps to maintain healthy boundaries between these family subsystems. Furthermore, parents could be helped to recognise the effect that one relationship, such as with their child with ID, has on their other relationships, particularly with their spouse or partner. There can be a tendency to target intervention only at the child with ID, or the parent-child relationship, however these results suggest that supporting family subsystems that do not just include children, such as the marital/partner subsystem, could be as important for the outcomes of all family members in two-parent families.

Emerging evidence from research on single mothers of children with ID in Chapter 5 which demonstrated the negative association between the impact of caregiving for a child with ID and psychological distress, indicates the requirement for increased provision which seeks to reduce the impact of caregiving on single mothers. There is a clear need for more formal sources of support and funding to ensure that single mothers are able to regularly engage in social and leisure activities and assume roles other than that of parent. Single mothers may not have the support from another caregiver or extended family member and so greater respite provision is likely to be welcomed. It was also apparent from this analysis that the positive gain associated with

parenting a child with ID need to be highlighted and harnessed because of their potential to improve single mothers' satisfaction with life.

Lastly, the findings indicate that mothers and fathers of children with ID may require different types and amounts of support. As shown in Chapter 2, mothers were more likely to report greater symptoms of depression compared to fathers. While this may be an issue of fathers under-reporting depressive symptoms, this finding is in line with previous research which suggests that the effects of parenting a child with ID are more severely experienced by mothers, which may be linked to their role as the primary caregiver for the child with ID. Furthermore, in Chapter 3, we have learnt that there were no differences between fathers of children with and without a child with ID on psychological wellbeing outcomes when controlling for a range of other factors – a finding which is in contrast to studies with mothers of children with ID. While mothers may benefit from additional help with caregiving responsibilities, this may not be the case for fathers. Poverty was a significant predictor of father wellbeing, therefore supporting fathers in most need may be the best way forward. Overall there is a clear need to find ways to engage with fathers of children with ID both in research and clinical practice in order to better understand their needs.

### **Limitations and Future Research**

This thesis has contributed new knowledge to the field of ID family research, however there are limitations which need to be discussed. Given the lack of research in all of the areas covered in the four empirical chapters, it is important to say that these analyses are a starting point and so further work with a broader range of variables is required. This thesis had a micro-level focus, and so future research should also consider variables within systems outside of the family unit and at the macro-level.

There is the potential for future work to conduct longitudinal analyses in order to capture families' outcomes across the lifecycle and allow for the exploration of causal

pathways. Future work should look to explore how the relationship satisfaction of parents of children with ID/ASD evolves over time as this was not feasible in the current thesis due to the lack of available longitudinal data. Furthermore, there may now be scope to examine father wellbeing in multiple waves of the MCS as this was not desirable in the current thesis due to the lack of existing cross-sectional work on paternal wellbeing in families of children with ID. It is also worth considering whether there are other data collection methods aside from surveys which can capture family functioning over time, such as diary studies which have already been used within the field.

The use of self-report survey data also means that the findings presented in this thesis could reflect perceptions and not necessarily reality. Furthermore, while the thesis used data that referred to different individuals or subsystems, most of the studies still relied on one source. Findings may be different if all parts of the system contribute their own data. Source variance is one of the main limitations of the primary data collected for the Cerebra 1000 Families study (which informed Chapters 4 and 5 of this thesis). Whilst the intention was to recruit two caregivers per family, mainly a mother and a father, the study was not successful in recruiting enough secondary caregivers and so these analyses are based on mothers who provided information about themselves and their family.

While it is widely acknowledged that fathers are more difficult to engage in research than mothers (Cassano, Adrian, Veits & Zeman, 2006; Phares, Fields, Kamboukos, & Lopez, 2005), such a low level of engagement was not expected given that fathers of children with ID were actively targeted and families were asked to complete (where possible) a primary and secondary caregiver survey. There are a number of suggestions as to why fathers may not participate as readily in research as mothers. Some indicate that studies may struggle to recruit fathers if they are not invited directly (Phares et al., 2005) or the importance of their participation is not explicitly emphasised (Costigan and Cox, 2001). While fathers were directly targeted in the Cerebra 1000 Families study via our on- and off-line recruitment streams it may still be

the case that these calls did not reach fathers if it was the mother who provided the email address and signed-up to mailing lists, or mothers being the ones who follow online feeds and attend events related to their child's disability. Phares et al. (2005) suggest that fathers may need alternative recruitment strategies, including contacting fathers through their place of work. It may also be that a study aimed at families was not specific enough to signify that we wanted fathers to participate. It has also been proposed that fewer fathers participate in research studies about their child because of the longstanding exclusion of fathers in matters which are related to their child's needs and provision (Phares, 1992) and the assumption that fathers are less present in the lives of their children (Phares, 1996). These assumptions may mean that fathers feel that they are not the 'best' person to provide information on their child, or that their experiences of parenting are of less value to research. Phares (1992) also describes how often there is an erroneous expectation that fathers are not available to take part in research due to work commitments, despite evidence to suggest that their participation is not restricted by employment (Costigan and Cox, 2001).

From reviewing the overall demographic of participants who took part in the 1000 Families study, it is evident that there were more two-parent families than any other family type and so more work needs to be done to investigate why other family members such as fathers and extended family members are not as willing to take part in research. It could be that having very distinct parallel streams of recruitment which directly target each family member could be more successful in obtaining reports from other members of the family unit. It is also evident that more direct reports from siblings and the child with ID are warranted, as their voices are still largely absent in ID research (Hastings, 2016).

Further limitations of the Cerebra 1000 Families study are that it is not population-representative and therefore does not reflect the experiences of all families in the UK. However, the study did not aim to be representative of families of children with ID in the UK but to be the largest study of its kind. There may also be sample biases

given the collaboration with the charity Cerebra as they are funders of the study and aided in the recruitment of participants, however it is also the case that their database of members that was used to distribute information about the study is UK-wide and that the team recruited via a number of other steams aside from that in partnership with Cerebra (as described in Chapter 1, p.16). Future research should continually strive to recruit a more representative sample of participants and engage with hard-to-reach caregivers, however this is a task which is significantly constrained by time and resources.

## **Conclusions**

This thesis contributes new knowledge to the field. It has provided a greater understanding of the interrelatedness of individual and subsystem functioning, and the relationship between subsystems and overall family functioning in families of children with ID, and has examined neglected parts and members of the family system, such as the marital/partner subsystem, and the outcomes of fathers and single mothers of children with ID. Overall the thesis argues that the families of children with ID matter. The functioning of the family unit and the wellbeing of its members has the power to shape the outcomes of the child with ID and so supporting families to thrive is important.

Research and clinical intervention which embraces the complexity of family dynamics and relationships will go further in improving the lives of children with ID and members of their family unit because of its ability to be applied in real-life contexts. Systemic relationships are a challenge to research, requiring sophisticated statistical techniques which often create more questions than answers. However, they are necessary in order to move on from simplistic notions of what raising a child with ID is like. This thesis does not wish to in any way downplay the many challenges that families of children with ID face, but it does ask for researchers and practitioners to engage with a narrative which accepts that family life and relationships are complex,

and as such there are a myriad of factors within the family system which can influence the experiences and outcomes of families of children with ID.

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## Appendices

**Appendix A: Ethical approval confirmation****NRES Committee West Midlands - South Birmingham**

Royal Standard Place  
Nottingham NG1 6FS  
Tel: 0115 883 9428

11 September 2015

Professor Richard Hastings  
CEDAR (Centre for Educational Development Appraisal and Research)  
University of Warwick  
Coventry UK  
CV47AL

Dear Professor Hastings

<b>Study title:</b>	<b>The 1,000 families study: Well-being in families of children with intellectual disability</b>
<b>REC reference:</b>	<b>15/WM/0267</b>
<b>IRAS project ID:</b>	<b>169882</b>

Thank you for your letter of 07 September 2015, responding to the Committee's request for further information on the above research and submitting revised documentation.

The further information has been considered on behalf of the Committee by the Chair.

We plan to publish your research summary wording for the above study on the HRA website, together with your contact details. Publication will be no earlier than three months from the date of this favourable opinion letter. The expectation is that this information will be published for all studies that receive an ethical opinion but should you wish to provide a substitute contact point, wish to make a request to defer, or require further information, please contact the REC Assistant, Nicola Kohut, [nrescommittee.westmidlands-southbirmingham@nhs.net](mailto:nrescommittee.westmidlands-southbirmingham@nhs.net). Under very limited circumstances (e.g. for student

research which has received an unfavourable opinion), it may be possible to grant an exemption to the publication of the study.

### **Confirmation of ethical opinion**

On behalf of the Committee, I am pleased to confirm a favourable ethical opinion for the above research on the basis described in the application form, protocol and supporting documentation as revised, subject to the conditions specified below.

### **Conditions of the favourable opinion**

The favourable opinion is subject to the following conditions being met prior to the start of the study.

Management permission or approval must be obtained from each host organisation prior to the start of the study at the site concerned.

*Management permission ("R&D approval") should be sought from all NHS organisations involved in the study in accordance with NHS research governance arrangements.*

Guidance on applying for NHS permission for research is available in the Integrated Research Application System or at <http://www.rdforum.nhs.uk>.

*Where a NHS organisation's role in the study is limited to identifying and referring potential participants to research sites ("participant identification centre"), guidance should be sought from the R&D office on the information it requires to give permission for this activity.*

*For non-NHS sites, site management permission should be obtained in accordance with the procedures of the relevant host organisation.*

*Sponsors are not required to notify the Committee of approvals from host organisations* Registration of Clinical Trials

All clinical trials (defined as the first four categories on the IRAS filter page) must be registered on a publically accessible database. This should be before the first participant is recruited but no later than 6 weeks after recruitment of the first participant.

There is no requirement to separately notify the REC but you should do so at the earliest opportunity e.g. when submitting an amendment. We will audit the registration details as part of the annual progress reporting process.

To ensure transparency in research, we strongly recommend that all research is registered but for non-clinical trials this is not currently mandatory.

If a sponsor wishes to request a deferral for study registration within the required timeframe, they should contact [hra.studyregistration@nhs.net](mailto:hra.studyregistration@nhs.net). The expectation is that all clinical trials will be registered, however, in exceptional circumstances non registration may be permissible with prior agreement from the HRA. Guidance on where to register is provided on the HRA website.

**It is the responsibility of the sponsor to ensure that all the conditions are complied with before the start of the study or its initiation at a particular site (as applicable).**

### **Ethical review of research sites**

#### **NHS sites**

The favourable opinion applies to all NHS sites taking part in the study, subject to management permission being obtained from the NHS/HSC R&D office prior to the start of the study (see "Conditions of the favourable opinion" below).

### **Approved documents**

The final list of documents reviewed and approved by the Committee is as follows:

<i>Document</i>	<i>Version</i>	<i>Date</i>
Copies of advertisement materials for research participants [Project short advertisement text]	1	13 July 2015
Evidence of Sponsor insurance or indemnity (non NHS Sponsors only) [Warwick University insurance 15-16]		
IRAS Checklist XML [Checklist_07092015]		07 September 2015
Letter from sponsor [Confirmation of Sponsorship]		
Other [CV Mikeda Jess]		
Other [CV Jane Margetson]		
Other [Elizabeth Halstead CV]		
Other [Response to initial ethics opinion]		
Participant consent form [Primary parental caregiver consent form]	1.1	03 September 2015
Participant consent form [Secondary Parental Caregiver consent form]	1.1	03 September 2015
Participant information sheet (PIS) [Primary caregiver information sheet]	1.1	03 September 2015
Participant information sheet (PIS) [Secondary caregiver information sheet]	1.1	03 September 2015
REC Application Form [REC_Form_14072015]		14 July 2015
Research protocol or project proposal [1000 Families study]	1	09 July 2015
Summary CV for Chief Investigator (CI) [Richard Hastings CV]		
Summary CV for student [CV Emma Langley]		
Summary CV for supervisor (student research) [CV Vaso Totsika]		
Validated questionnaire [Developmental Behaviour Checklist (DBC)]		
Validated questionnaire [Online survey items and questionnaires]	1.1	03 September 2015

## Statement of compliance

The Committee is constituted in accordance with the Governance Arrangements for Research Ethics Committees and complies fully with the Standard Operating Procedures for Research Ethics Committees in the UK.

## After ethical review

### Reporting requirements

The attached document “*After ethical review – guidance for researchers*” gives detailed guidance on reporting requirements for studies with a favourable opinion, including:

- Notifying substantial amendments
  - Adding new sites and investigators
  - Notification of serious breaches of the protocol
  - Progress and safety reports
  - Notifying the end of the study
- The HRA website also provides guidance on these topics, which is updated in the light of changes in reporting requirements or procedures.

## User Feedback

The Health Research Authority is continually striving to provide a high quality service to all applicants and sponsors. You are invited to give your view of the service you have received and the application procedure. If you wish to make your views known please use the feedback form available on the HRA website: <http://www.hra.nhs.uk/about-the-hra/governance/quality-assurance/>

## HRA Training

We are pleased to welcome researchers and R&D staff at our training days – see details at <http://www.hra.nhs.uk/hra-training/>

## **15/WM/0267 Please quote this number on all correspondence**

With the Committee’s best wishes for the success of this project.

Yours Sincerely,

PP: 

**Professor Simon Bowman**  
**Chair**

Email: nrescommittee.westmidlands-southbirmingham@nhs.net

*Enclosures:* After ethical review – guidance for researchers

*Copy to:* Mrs Jane Prewett

## Appendix B: Ethical approval protocol

IRAS Version 4.0.0

Welcome to the Integrated Research Application System

### IRAS Project Filter

The integrated dataset required for your project will be created from the answers you give to the following questions. The system will generate only those questions and sections which (a) apply to your study type and (b) are required by the bodies reviewing your study. Please ensure you answer all the questions before proceeding with your applications.

Please complete the questions in order. If you change the response to a question, please select 'Save' and review all the questions as your change may have affected subsequent questions.

**Please enter a short title for this project** (maximum 70 characters)

The 1,000 Families Study

**1. Is your project research?**

☒ Yes ☐ No

**2. Select one category from the list below:**

- ☐ Clinical trial of an investigational medicinal product
- ☐ Clinical investigation or other study of a medical device
- ☐ Combined trial of an investigational medicinal product and an investigational medical device
- ☐ Other clinical trial to study a novel intervention or randomised clinical trial to compare interventions in clinical practice
- ☐ Basic science study involving procedures with human participants
- ☒ Study administering questionnaires/interviews for quantitative analysis, or using mixed quantitative/qualitative methodology
- ☐ Study involving qualitative methods only
- ☐ Study limited to working with human tissue samples (or other human biological samples) and data (specific project only)
- ☐ Study limited to working with data (specific project only)
- ☐ Research tissue bank
- ☐ Research database

**If your work does not fit any of these categories, select the option below:**

☐ Other study

**2a. Please answer the following question(s):**

- a) Does the study involve the use of any ionising radiation? ☐ Yes ☒ No
- b) Will you be taking new human tissue samples (or other human biological samples)? ☐ Yes ☒ No
- c) Will you be using existing human tissue samples (or other human biological samples)? ☐ Yes ☒ No

**3. In which countries of the UK will the research sites be located?** (Tick all that apply)

- ☒ England
- ☒ Scotland
- ☒ Wales
- ☒ Northern Ireland



NHS REC Form

Reference:  
15/WM/0267

IRAS Version 4.0.0

**3a. In which country of the UK will the lead NHS R&D office be located:**

- ☐ England  
☐ Scotland  
☐ Wales  
☐ Northern Ireland  
☒ This study does not involve the NHS

**4. Which review bodies are you applying to?**

- ☐ NHS/HSC Research and Development offices  
☐ Social Care Research Ethics Committee  
☒ Research Ethics Committee  
☐ Confidentiality Advisory Group (CAG)  
☐ National Offender Management Service (NOMS) (Prisons & Probation)

*For NHS/HSC R&D offices, the CI must create Site-Specific Information Forms for each site, in addition to the study-wide forms, and transfer them to the PIs or local collaborators.*

**5. Will any research sites in this study be NHS organisations?**

- ☐ Yes    ☒ No

**6. Do you plan to include any participants who are children?**

- ☐ Yes    ☒ No

**7. Do you plan at any stage of the project to undertake intrusive research involving adults lacking capacity to consent for themselves?**

- ☐ Yes    ☒ No

*Answer Yes if you plan to recruit living participants aged 16 or over who lack capacity, or to retain them in the study following loss of capacity. Intrusive research means any research with the living requiring consent in law. This includes use of identifiable tissue samples or personal information, except where application is being made to the Confidentiality Advisory Group to set aside the common law duty of confidentiality in England and Wales. Please consult the guidance notes for further information on the legal frameworks for research involving adults lacking capacity in the UK.*

**8. Do you plan to include any participants who are prisoners or young offenders in the custody of HM Prison Service or who are offenders supervised by the probation service in England or Wales?**

- ☐ Yes    ☒ No

**9. Is the study or any part of it being undertaken as an educational project?**

- ☒ Yes    ☐ No

Please describe briefly the involvement of the student(s):

Three PhD students will work on data collection for this survey. Each will be examining and reporting data from a different area of the survey, with their own research questions, to contribute towards their PhD theses.

**9a. Is the project being undertaken in part fulfilment of a PhD or other doctorate?**

- ☒ Yes    ☐ No

**10. Will this research be financially supported by the United States Department of Health and Human Services or any of its divisions, agencies or programs?**

☐ Yes ☒ No

**11. Will identifiable patient data be accessed outside the care team without prior consent at any stage of the project (including identification of potential participants)?**

☐ Yes ☒ No

DRAFT

**Integrated Research Application System****Application Form for Research administering questionnaires/interviews for quantitative analysis or mixed methodology study****Application to NHS/HSC Research Ethics Committee**

The Chief Investigator should complete this form. Guidance on the questions is available wherever you see this symbol displayed. We recommend reading the guidance first. The complete guidance and a glossary are available by selecting [Help](#).

Please define any terms or acronyms that might not be familiar to lay reviewers of the application.

**Short title and version number:** (maximum 70 characters - this will be inserted as header on all forms)  
The 1,000 Families Study

*Please complete these details after you have booked the REC application for review.*

**REC Name:**  
West Midlands - South Birmingham

**REC Reference Number:**  
15/WM/0267

**Submission date:**  
14/07/2015

**PART A: Core study information****1. ADMINISTRATIVE DETAILS****A1. Full title of the research:**

The 1,000 families study: Well-being in families of children with intellectual disability

**A2-1. Educational projects**

Name and contact details of student(s):

**Student 1**

	Title Forename/Initials Surname
	Miss Emma Langley
Address	CEDAR (Centre for Educational Development Appraisal and Research)
	University of Warwick
	Coventry UK
Post Code	CV47AL
E-mail	e.langley@warwick.ac.uk
Telephone	07505171776
Fax	

Give details of the educational course or degree for which this research is being undertaken:

Name and level of course/ degree:  
PhD, Education and Psychology

Name of educational establishment:  
University of Warwick

#### Student 2

	Title	Forename/Initials	Surname
	Miss	Jane	Margetson
Address	CEDAR (Centre for Educational Development Appraisal and Research)		
	University of Warwick		
	Coventry UK		
Post Code	CV47AL		
E-mail	J.Margetson@Warwick.ac.uk		
Telephone	07891556975		
Fax			

Give details of the educational course or degree for which this research is being undertaken:

Name and level of course/ degree:  
PhD, Education and Psychology

Name of educational establishment:  
University of Warwick

#### Student 3

	Title	Forename/Initials	Surname
	Miss	Mikeda	Jess
Address	CEDAR (Centre for Educational Development Appraisal and Research)		
	University of Warwick		
	Coventry UK		
Post Code	CV47AL		
E-mail	m.jess@warwick.ac.uk		
Telephone			
Fax			

Give details of the educational course or degree for which this research is being undertaken:

Name and level of course/ degree:  
PhD, Education and Psychology

Name of educational establishment:  
University of Warwick

Name and contact details of academic supervisor(s):

#### Academic supervisor 1

	Title	Forename/Initials	Surname
	Professor	Richard	Hastings
Address	CEDAR (Centre for Educational Development Appraisal and Research)		

NHS REC Form

Reference:  
15/WM/0267

IRAS Version 4.0.0

University of Warwick  
Coventry UK  
Post Code CV47AL  
E-mail r.hastings@warwick.ac.uk  
Telephone 02476 522197  
Fax

**Academic supervisor 2**

Title Forename/Initials Surname  
Dr Vaso Totsika  
Address CEDAR (Centre for Educational Development Appraisal and Research)  
University of Warwick  
Coventry UK  
Post Code CV4 7AL  
E-mail V.Totsika@warwick.ac.uk  
Telephone 024 765 22185  
Fax

Please state which academic supervisor(s) has responsibility for which student(s):

Please click "Save now" before completing this table. This will ensure that all of the student and academic supervisor details are shown correctly.

Student(s)	Academic supervisor(s)
<b>Student 1</b> Miss Emma Langley	<input checked="" type="checkbox"/> Professor Richard Hastings <input checked="" type="checkbox"/> Dr Vaso Totsika
<b>Student 2</b> Miss Jane Margetson	<input checked="" type="checkbox"/> Professor Richard Hastings <input checked="" type="checkbox"/> Dr Vaso Totsika
<b>Student 3</b> Miss Mikeda Jess	<input checked="" type="checkbox"/> Professor Richard Hastings <input checked="" type="checkbox"/> Dr Vaso Totsika

A copy of a current CV for the student and the academic supervisor (maximum 2 pages of A4) must be submitted with the application.

**A2-2. Who will act as Chief Investigator for this study?**

- ☐ Student  
☒ Academic supervisor  
☐ Other

**A3-1. Chief Investigator:**

Title Forename/Initials Surname  
Professor Richard Hastings  
Post Cerebra Chair of Family Research  
Qualifications BSc, PhD, CPsychol

NHS REC Form

Reference:  
15/WM/0267

IRAS Version 4.0.0

Employer	University of Warwick
Work Address	CEDAR (Centre for Educational Development Appraisal and Research) University of Warwick Coventry UK
Post Code	CV47AL
Work E-mail	r.hastings@warwick.ac.uk
* Personal E-mail	
Work Telephone	+44 (0)2476 522197
* Personal Telephone/Mobile	
Fax	

*\* This information is optional. It will not be placed in the public domain or disclosed to any other third party without prior consent.  
A copy of a current CV (maximum 2 pages of A4) for the Chief Investigator must be submitted with the application.*

**A4. Who is the contact on behalf of the sponsor for all correspondence relating to applications for this project?**  
This contact will receive copies of all correspondence from REC and R&D reviewers that is sent to the CI.

	Title Forename/Initials Surname
	Mrs Jane Prewett
Address	Research Impact Services Coventry
Post Code	CV4 8UW
E-mail	WMSSponsorship@warwick.ac.uk
Telephone	02476522746
Fax	

**A5-1. Research reference numbers.** Please give any relevant references for your study:

Applicant's/organisation's own reference number, e.g. R & D (if available):	R.CDAA.3049
Sponsor's/protocol number:	
Protocol Version:	1.0
Protocol Date:	12/06/2015
Funder's reference number:	N/A
Project website:	

**Additional reference number(s):**

Ref.Number Description	Reference Number
Sponsor reference number	REGO-2015-1604

*Registration of research studies is encouraged wherever possible. You may be able to register your study through your NHS organisation or a register run by a medical research charity, or publish your protocol through an open access publisher. If you have registered your study please give details in the "Additional reference number(s)" section.*

**A5-2. Is this application linked to a previous study or another current application?**

☐ Yes ☒ No

*Please give brief details and reference numbers.*

**2. OVERVIEW OF THE RESEARCH**

*To provide all the information required by review bodies and research information systems, we ask a number of specific questions. This section invites you to give an overview using language comprehensible to lay reviewers and members of the public. Please read the guidance notes for advice on this section.*

**A6-1. Summary of the study.** Please provide a brief summary of the research (maximum 300 words) using language easily understood by lay reviewers and members of the public. Where the research is reviewed by a REC within the UK Health Departments Research Ethics Service, this summary will be published on the website of the National Research Ethics Service following the ethical review.

What are the experiences of UK families with a child with an intellectual disability?

This large-scale longitudinal study investigates the impact on families when they have a child with an intellectual disability. The research is funded by the research charity Cerebra and will recruit 1000 or more families in the UK with a child with an intellectual disability aged 4-11 years. Families will be recruited in a range of ways including social media, communication with special schools, and support from third sector organisations. Families will be asked to participate in Wave 1 of the study (2015-2017) and indicate whether they would also like to be contacted in two years time for Wave 2 (2017-2019).

Wave 1 of the study asks primary and secondary caregivers within each family to report on many aspects of their lives including their own health, well-being and life satisfaction; their children's behaviour and relationships, and general family functioning. The majority of this information will be collected by a self-report online survey, with additional information about the child with the intellectual disability's behaviour collected via a telephone interview (with the primary caregiver).

There is no existing study of this type and size in the UK and so this research will make a significant leap forward in intellectual disability family research and has the potential to lead to decades of high quality family research. Crucially, the outcomes of this research will be important for supporting families. Input from the research charity Cerebra, means that the findings will be made accessible for families and practitioners and will be regularly disseminated at different time points throughout the study.

**A6-2. Summary of main issues.** Please summarise the main ethical, legal, or management issues arising from your study and say how you have addressed them.

*Not all studies raise significant issues. Some studies may have straightforward ethical or other issues that can be identified and managed routinely. Others may present significant issues requiring further consideration by a REC, R&D office or other review body (as appropriate to the issue). Studies that present a minimal risk to participants may raise complex organisational or legal issues. You should try to consider all the types of issues that the different reviewers may need to consider.*

The physical and ethical risks to the researcher and the participant in this project are minimal because of the use of an online survey method. However, the team is aware that there is a small chance that the survey could cause some emotional distress for caregivers, so have made sure that this is documented in any information prior to consent. As the researcher will not be present when the survey is being completed, it has been ensured that participants know who they can contact if they experience any discomfort or have any concerns. It will be made very clear that they can decide to not complete the survey, or withdraw themselves and any data that they have provided from the study without any reason.

We are also aware that completing the survey will be an additional burden for families. We have tried to negate this in three ways: firstly, by making the survey as short as possible, using essential measures (while still trying to retain the items that will answer the research questions). Secondly, we decided to split the surveys into one online survey and one telephone interview (the interview will only need to be completed by one caregiver if they are willing to do so). Thirdly, participants can fill in the online survey at their own leisure and can opt out of the telephone interview or arrange to complete it at a time that is suitable for them.

A challenge for the project will be to recruit approximately 1000 families, thus the research team has decided to recruit via a number of streams including information on a project website, social media and contact with schools and third sector organisations. Participants will be able to opt-in to the study by clicking through to the survey via the link provided on the study advertisement. If they then decide then that they do not want to proceed, they are under no obligation to continue.

Due to the large-scale nature of the study it is not feasible to adapt the survey or make it available in a language other

than English because of time and cost considerations. While making it available in other languages and mediums is desirable, it would not be possible to adapt the survey for each individual. Additionally, translating existing standardised assessment measures may be difficult due to copyright considerations.

There should be no significant issues for obtaining informed consent because the providing consent is a condition for taking part in the study. Nevertheless we will be providing potential participants with clear yet detailed information on the participation page online. We will make it clear that participants can opt into all stages of the research, including being contacted in two years' time to participate in wave 2 of the project.

Keeping the personal data of participants secure is a key consideration of this study, as the study will be obtaining a wealth of personal information from families including contact and demographic information. This information will only be available to the research team. The electronic data gathered on participants, including personal/demographic information, will be stored securely on firewalled university computers.

The research team will consider the confidentiality of participant data by separating respondents' contact information from their survey responses. These two types of information will be stored on separate secure databases. No individual reporting of results will be provided and practically would not be viable because of the size of the study. Confidentiality will only be broken if a respondent reports something which suggests that they or someone they care for is at risk, and therefore appropriate authorities would be notified. This information will be made clear in the consent procedure.

Any dissemination of results through reports journal articles, conferences or PhD theses will be based on the population of respondents as a whole.

Participants will be made aware that they can view updates and reports from the study via the project website and Facebook and Twitter pages.

**A6-3. Proportionate review of REC application** *The initial project filter has identified that your study may be suitable for proportionate review by a REC sub-committee. Please consult the current guidance notes from NRES and indicate whether you wish to apply through the proportionate review service or, taking into account your answer to A6-2, you consider there are ethical issues that require consideration at a full REC meeting.*

☐ Yes - proportionate review ☒ No - review by full REC meeting

*Further comments (optional):*

This study raises no significant medical issues, and we believe it is suitable for proportionate review. However, we are building a sample that is likely to be invited to participate in research several times into the future (after the currently proposed Wave 2). Therefore, we are suggesting that a full review would be appropriate.

*Note: This question only applies to the REC application.*

### 3. PURPOSE AND DESIGN OF THE RESEARCH

**A7. Select the appropriate methodology description for this research. Please tick all that apply.**

- ☐ Case series/ case note review
- ☐ Case control
- ☐ Cohort observation
- ☐ Controlled trial without randomisation
- ☐ Cross-sectional study
- ☐ Database analysis
- ☐ Epidemiology
- ☐ Feasibility/ pilot study
- ☐ Laboratory study
- ☐ Metanalysis
- ☐ Qualitative research
- ☒ Questionnaire, interview or observation study



- ☐ Randomised controlled trial
- ☐ Other (please specify)

**A10. What is the principal research question/objective? Please put this in language comprehensible to a lay person.**

We are not distinguishing between one main objective and secondary objectives.

The main aims of this research study are as follows:

1. To describe the well-being of family members of children with intellectual disability and to compare these data wherever possible with normative data and/or to national datasets (especially the Millennium Cohort Study). Family members include mothers (or primary parental caregivers), fathers (or secondary parental caregivers), and siblings.
2. To examine well-being differences between mothers and fathers in families of children with intellectual disability.
3. To explore correlates of maternal, paternal and sibling well-being, including: family deprivation; other child, parent, sibling and family demographic factors; other family members' well-being, and the child with intellectual disability's behaviour problems and adaptive skills.
4. To explore correlates of the behavioural and emotional well-being of the child with intellectual disability, including: family deprivation; other child, parent, sibling and family demographic factors; other family members' well-being, the child with intellectual disability's adaptive skills, and parenting attitudes and behaviours.

Using a repeated data collection, two years after the initial survey, we will also examine aims 3 and 4 with a focus on longitudinal associations.

**A11. What are the secondary research questions/objectives if applicable? Please put this in language comprehensible to a lay person.**

We are not distinguishing between one main objective and secondary objectives.

The main aims of this research study are as follows:

1. To describe the well-being of family members of children with intellectual disability and to compare these data wherever possible with normative data and/or to national datasets (especially the Millennium Cohort Study). Family members include mothers (or primary parental caregivers), fathers (or secondary parental caregivers), and siblings.
2. To examine well-being differences between mothers and fathers in families of children with intellectual disability.
3. To explore correlates of maternal, paternal and sibling well-being, including: family deprivation; other child, parent, sibling and family demographic factors; other family members' well-being, and the child with intellectual disability's behaviour problems and adaptive skills.
4. To explore correlates of the behavioural and emotional well-being of the child with intellectual disability, including: family deprivation; other child, parent, sibling and family demographic factors; other family members' well-being, the child with intellectual disability's adaptive skills, and parenting attitudes and behaviours.

Using a repeated data collection, two years after the initial survey, we will also examine aims 3 and 4 with a focus on longitudinal associations.

**A12. What is the scientific justification for the research? Please put this in language comprehensible to a lay person.**

As there has not been a study of this kind on such a large scale in the UK, the findings will be an original contribution to scientific knowledge. To date, intellectual disability (ID) and family research has primarily explored the negative impact of raising a child with ID; however the current study aims to take the position from a positive standpoint, whilst also allowing family members to include any potential negative aspects involved with raising their child. Furthermore, there is limited research on the experiences of the secondary caregivers of children with ID.

This study aims to fill that gap in the scientific literature. The potential findings have both theoretical and practical implications. This large-scale longitudinal study will have a significant impact on intellectual disability research in the UK, with the potential to lead to decades of high quality family research. In addition the findings could identify the key needs of families of children with ID which may lead to the development of practical support and interventions for these families. Thus this research has direct implications for both research and families with an ID child.

**A13. Please summarise your design and methodology. It should be clear exactly what will happen to the research participant, how many times and in what order. Please complete this section in language comprehensible to the lay person. Do not simply reproduce or refer to the protocol. Further guidance is available in the guidance notes.**

The large-scale nature of this study necessitates a quantitative design. A qualitative study with one or two home visits to 1,000 families would be prohibitively expensive in human resource and travel costs. The use of a quantitative design is building on previous research in this area which has been small-scale or qualitative in design.

The decision to use an online survey as the main method of data collection reflects the aim to collect vast amounts of data: it will allow for the data to be collected easily at fairly low cost and will reduce the time it takes to complete the research for participants and researchers. The survey also has other benefits in that the measures used can be completed swiftly and can be quantified with ease. Given that we are asking for the participation of families of children with a disability, we feel that the survey will not inconvenience them too much and can be completed in their own homes at their own leisure. The decision to have an online survey and then a telephone interview for the primary caregiver was made to prevent survey 1 becoming too long for the participant to complete. The measures used in the telephone interview can be rapidly completed.

#### Timetable for participants (Wave 1 only)

1. Participants will see/receive information about the study and can go to the online survey information via the link provided on the project advertisement.
2. Participants will be asked to indicate on the online survey page whether they are a primary or a secondary caregiver (some definition/guidance is provided) and this will direct them to the full study information and the consent page.
3. Participants will be required to answer all the consent questions, including whether they wish to be contacted for a telephone interview (Primary caregiver only) and for Wave 2 of the study in two years' time.
4. Once consent has been completed participants can proceed with the survey.
5. When a survey has been completed, a researcher will export the information from SurveyMonkey and will extract contact information and arrange a telephone interview with participants (Primary Caregivers only) who stated that they were happy to be contacted.
6. The telephone interview is then completed (Primary Caregiver only).
7. Participants who indicate that they would be willing to be invited into Wave 2 of the study will be contacted after approximately two years (by email, telephone or post).
8. If participants are happy to participate in Wave 2, the process will start again with the consent questions in the online survey.

If participants have indicated that would like to be kept informed about the results of this study and other family research carried out by the research group, they will be sent a regular e-newsletter. The same information will be available via social media if participants opt not to receive the newsletter.

As the research team will not be meeting any of the participants responding to the survey the influence of researcher bias will be minimal. Nevertheless, in designing the survey the research team have worked together to ensure that the survey is balanced in what it explores and that the measures used have been appraised prior to being included. The use of measures within the study also help to negate any researcher influence, in that they are closed questions and are reported either directly by the respondent online and for the telephone interview marked down on a standardised measure form. In the analysis stage, the research team will work together to analyse the data and PhD students will be guided by their supervisors who will check claims made in their reports and theses.

The main focus of the research is on a large sample to enable more complex research questions to be addressed, and on collecting data about multiple family members. An online survey is the most efficient and effective way to achieve these broader aims. We recognise that the sample obtained will not be representative of the UK population of parents of children with intellectual disability. Other research being carried out by our team addresses this issue of representativeness through secondary analysis of UK population-based datasets. A limitation of existing population-based surveys is the small number of families included who have children with intellectual disability. Thus, our research programme as a whole does address representativeness but this is not a priority for the 1,000 Families Study.

**A14-1. In which aspects of the research process have you actively involved, or will you involve, patients, service users, and/or their carers, or members of the public?**

☐ Design of the research

- ☐ Management of the research  
☐ Undertaking the research  
☐ Analysis of results  
☒ Dissemination of findings  
☐ None of the above

*Give details of involvement, or if none please justify the absence of involvement.*

We have secured initial Impact funding separate from the research grant to support the recruitment of up to 6 family members to become part of a parent advisory group for this project and to establish themselves as themselves as "Ambassadors" for the research. This role will involve sharing the results with other families, and we will be providing training/mentoring for this group of family members.

#### 4. RISKS AND ETHICAL ISSUES

##### RESEARCH PARTICIPANTS

##### A17-1. Please list the principal inclusion criteria (list the most important, max 5000 characters).

Families with one or more children aged between 4 and 11 years old who have an intellectual disability as reported by a parental caregiver.

Families with at least one parental caregiver responding to the on-line survey.

Families that live in the UK.

##### A17-2. Please list the principal exclusion criteria (list the most important, max 5000 characters).

Families where the child with an intellectual disability lives outside of the family home on a full time basis (e.g. a 52 week residential school placement).

Parental caregivers whose English literacy skills mean they are unable to participate in an on-line and telephone interview.

Parental caregivers under 18 years of age.

##### RESEARCH PROCEDURES, RISKS AND BENEFITS

##### A18. Give details of all non-clinical intervention(s) or procedure(s) that will be received by participants as part of the research protocol. These include seeking consent, interviews, non-clinical observations and use of questionnaires.

Please complete the columns for each intervention/procedure as follows:

- Total number of interventions/procedures to be received by each participant as part of the research protocol.
- If this intervention/procedure would be routinely given to participants as part of their care outside the research, how many of the total would be routine?
- Average time taken per intervention/procedure (minutes, hours or days)
- Details of who will conduct the intervention/procedure, and where it will take place.

Intervention or procedure	1	2	3	4
Consent form	1	0	5 minutes	The participants will complete the consent form as the first section of the online survey. The participant will need to consent before they can continue.
Online Survey	13	0	30-40 minutes	Participants will complete the survey online. 13 different sections are included - demographic questions, and then research measures as specified in the protocol.

Telephone interview	20	35-75 minutes	Education and Psychology PhD students based at the University of Warwick will conduct the telephone interviews from their private office. Participants will be instructed to be at in a private place (e.g. a room away from the family/colleagues/other distractions) when the telephone interview takes place. Only the primary caregiver will complete the telephone interview.
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**A21. How long do you expect each participant to be in the study in total?**

4 years

**A22. What are the potential risks and burdens for research participants and how will you minimise them?**

*For all studies, describe any potential adverse effects, pain, discomfort, distress, intrusion, inconvenience or changes to lifestyle. Only describe risks or burdens that could occur as a result of participation in the research. Say what steps would be taken to minimise risks and burdens as far as possible.*

The physical risks to participants are minimal as the research questions are answered via an online survey and telephone interview.

The survey and interview has a very small risk of leading to distress in the participant. Participants will be informed about the nature of the survey/interview before they take part so they are prepared for any feelings that might arise. Before participating, individuals will also be informed of the procedures for contacting the lead researcher/sponsor within a reasonable time, if following participation they experience stress, harm or have any concerns.

The time it will take for individuals to participate in this study may be an additional burden/inconvenience to families of children with an intellectual disability, on top of their caring/other responsibilities. Due care has been given to keep the number of research questions to a minimum and for participants to complete the survey and interview at different times (that suit them), to ensure participation is not overly burdensome.

To minimise these risks we will:

- i) Ensure informed consent
- ii) Ensure PhD students managing the study are supervised and supported appropriately.

**A23. Will interviews/ questionnaires or group discussions include topics that might be sensitive, embarrassing or upsetting, or is it possible that criminal or other disclosures requiring action could occur during the study?**

☐ Yes ☒ No

**A24. What is the potential for benefit to research participants?**

The only benefits for participants are the opportunity to contribute to increased understanding of families of children with intellectual disability, and thus to ensure more appropriately designed support services and interventions, and to receive accessible information about the study findings and similar research from our research group and others.

**A26. What are the potential risks for the researchers themselves? (if any)**

There are no risks to researchers themselves as the survey is conducted online and the interviews are by telephone.

**RECRUITMENT AND INFORMED CONSENT**

*In this section we ask you to describe the recruitment procedures for the study. Please give separate details for different study groups where appropriate.*

**A27-1. How will potential participants, records or samples be identified? Who will carry this out and what resources will be used? For example, identification may involve a disease register, computerised search of GP records, or review of medical records. Indicate whether this will be done by the direct healthcare team or by researchers acting under arrangements with the responsible care organisation(s).**

Participants will be recruited by PhD researchers in the Family Research team. Time and resourcing as part of their PhD is available for them to fulfil this task.

Information about the study will be disseminated through social media activities, such as a project website, Facebook page, and Twitter feed.

Special schools will be contacted and asked to forward information about the study to caregivers.

Recruitment will also be supported by third sector organisations, distributing information through their social media presence and regular newsletters.

**A27-2. Will the identification of potential participants involve reviewing or screening the identifiable personal information of patients, service users or any other person?**

☐ Yes ☒ No

*Please give details below:*

**A28. Will any participants be recruited by publicity through posters, leaflets, adverts or websites?**

☒ Yes ☐ No

*If Yes, please give details of how and where publicity will be conducted, and enclose copy of all advertising material (with version numbers and dates).*

A short advert describing the project will be published on-line through a project website and dedicated project Facebook and Twitter site. The same short text will be used by other organisations bringing the study to the attention of families.

**A29. How and by whom will potential participants first be approached?**

No individual participants will be directly approached by a member of the research team to take part in the study. Individuals who have seen information about the study online or have been directed to the online advert by special schools or third sector organisations will be able to go online to view the full information about the study and participate in the study if they so choose.

**A30-1. Will you obtain informed consent from or on behalf of research participants?**

☒ Yes ☐ No

*If you will be obtaining consent from adult participants, please give details of who will take consent and how it will be done, with details of any steps to provide information (a written information sheet, videos, or interactive material). Arrangements for adults unable to consent for themselves should be described separately in Part B Section 6, and for children in Part B Section 7.*

*If you plan to seek informed consent from vulnerable groups, say how you will ensure that consent is voluntary and fully informed.*

The advertisement for the study will provide a link to the first page of the online survey where full information about involvement will be available. If caregivers are happy to participate they can proceed to the consent page where they will be required to read and agree to a series of consent statements before proceeding.

Caregivers will not be able to start the survey if they have not answered the consent questions.

If caregivers have any questions about the project, they will be advised to contact the research team via the project email address provided on the information page.

*If you are not obtaining consent, please explain why not.*

*Please enclose a copy of the information sheet(s) and consent form(s).*

**A30-2. Will you record informed consent (or advice from consultees) in writing?**

☐ Yes ☒ No

*If No, how will it be recorded?*

There will be an electronic record created as a part of the response to the online survey rather than a physical signed consent form.

**A31. How long will you allow potential participants to decide whether or not to take part?**

There will be no specific time frame imposed on participants to decide whether they take part in the study. They can read the online information about the study at their own leisure and without any commitment to participate.

We anticipate that the survey will be live for between 12 and 18 months.

**A33-1. What arrangements have been made for persons who might not adequately understand verbal explanations or written information given in English, or who have special communication needs?(e.g. translation, use of interpreters)**

Caregivers who cannot complete the study because of their English literacy skills or special communication needs will not be involved in the study.

**A33-2. What arrangements will you make to comply with the principles of the Welsh Language Act in the provision of information to participants in Wales?**

The online and telephone survey will be provided in English only.

**A35. What steps would you take if a participant, who has given informed consent, loses capacity to consent during the study? Tick one option only.**

- ☐ The participant and all identifiable data or tissue collected would be withdrawn from the study. Data or tissue which is not identifiable to the research team may be retained.
- ☐ The participant would be withdrawn from the study. Identifiable data or tissue already collected with consent would be retained and used in the study. No further data or tissue would be collected or any other research procedures carried out on or in relation to the participant.
- ☐ The participant would continue to be included in the study.
- ☐ Not applicable – informed consent will not be sought from any participants in this research.
- ☒ Not applicable – it is not practicable for the research team to monitor capacity and continued capacity will be assumed.

*Further details:*

Due to the large-scale nature of the study, and the non-clinical nature of the sample, the research team will not be able to monitor capacity and therefore will assume continued capacity unless notified otherwise.

**CONFIDENTIALITY**

In this section, personal data means any data relating to a participant who could potentially be identified. It includes pseudonymised data capable of being linked to a participant through a unique code number.

**Storage and use of personal data during the study****A36. Will you be undertaking any of the following activities at any stage (including in the identification of potential participants)?(Tick as appropriate)**

- ☐ Access to medical records by those outside the direct healthcare team

- ☒ Electronic transfer by magnetic or optical media, email or computer networks
- ☐ Sharing of personal data with other organisations
- ☐ Export of personal data outside the EEA
- ☒ Use of personal addresses, postcodes, faxes, emails or telephone numbers
- ☐ Publication of direct quotations from respondents
- ☐ Publication of data that might allow identification of individuals
- ☐ Use of audio/visual recording devices
- ☒ Storage of personal data on any of the following:
  - ☐ Manual files including X-rays
  - ☐ NHS computers
  - ☒ Home or other personal computers
  - ☒ University computers
  - ☐ Private company computers
  - ☒ Laptop computers

*Further details:*

Researchers will use email as one of the media to communicate with participants, using email addresses that the parents have provided - to answer questions about the study, to make arrangements for telephone interviews, and to disseminate study results to parents. Emails with parents will not be used as a means to send or receive personal data.

Participants personal information including names and contact information is required to contact participants for the follow up study if they have agreed to this on the consent form, and to ensure that we can match together parents from the same family (required for analysis).

If participants would like to be kept up-to-date with the research findings we will also use their contact details to do this. Participants' telephone numbers will also be used if they have agreed to participate in the telephone interview stage of the study.

Personal data will be stored electronically on fire-walled University computers. These computers will be password protected and only accessible by the named researchers and the Chief Investigator.

Anonymised data only will be downloaded to personal computers or laptops for the purposes of data analysis. Data for analysis will include identifying participant numbers but the database linking these numbers to personal information will not be downloaded or transferred to personal or laptop computers. All personal and laptop computers will be password-protected as will the folders containing any data analysis file.

**A38. How will you ensure the confidentiality of personal data?** *Please provide a general statement of the policy and procedures for ensuring confidentiality, e.g. anonymisation or pseudonymisation of data.*

Participants will be assigned a randomised number to maintain anonymity.

Participants' responses will be pooled together with other participants for analyses so that that individual responses cannot be identified in any way.

Participants' names and personal information will not be able to be identified in any report or presentation about the study.

**A40. Who will have access to participants' personal data during the study?** *Where access is by individuals outside the direct care team, please justify and say whether consent will be sought.*

The named PhD researchers, (Mikeda Jess, Emma Langley and Jane Margetson) and their supervisors (Prof Richard Hastings and Dr Vaso Totsika), and part-time research assistant Liz Halstead.

**A43. How long will personal data be stored or accessed after the study has ended?**

- ☐ Less than 3 months  
☐ 3 – 6 months  
☐ 6 – 12 months  
☐ 12 months – 3 years  
☒ Over 3 years

*If longer than 12 months, please justify:*

Data collected for this study is to be used as part of an on going longitudinal study. Permission from participants will be granted in the consent procedure to be invited to participate in a follow-up survey 2 years from the initial survey.

**INCENTIVES AND PAYMENTS****A46. Will research participants receive any payments, reimbursement of expenses or any other benefits or incentives for taking part in this research?**

- ☐ Yes    ☒ No

**A47. Will individual researchers receive any personal payment over and above normal salary, or any other benefits or incentives, for taking part in this research?**

- ☐ Yes    ☒ No

**A48. Does the Chief Investigator or any other investigator/collaborator have any direct personal involvement (e.g. financial, share holding, personal relationship etc.) in the organisations sponsoring or funding the research that may give rise to a possible conflict of interest?**

- ☐ Yes    ☒ No

**NOTIFICATION OF OTHER PROFESSIONALS****A49-1. Will you inform the participants' General Practitioners (and/or any other health or care professional responsible for their care) that they are taking part in the study?**

- ☐ Yes    ☒ No

*If Yes, please enclose a copy of the information sheet/letter for the GP/health professional with a version number and date.*

**PUBLICATION AND DISSEMINATION****A50. Will the research be registered on a public database?**

- ☐ Yes    ☒ No

*Please give details, or justify if not registering the research.*

This study does not involve the trial of an intervention. Cerebra, as the funding organisation, will include information about the study and its progress on their website.

*Registration of research studies is encouraged wherever possible.*

*You may be able to register your study through your NHS organisation or a register run by a medical research charity.*



or publish your protocol through an open access publisher. If you are aware of a suitable register or other method of publication, please give details. If not, you may indicate that no suitable register exists. Please ensure that you have entered registry reference number(s) in question A5-1.

**A51. How do you intend to report and disseminate the results of the study? Tick as appropriate:**

- ☒ Peer reviewed scientific journals
- ☐ Internal report
- ☒ Conference presentation
- ☒ Publication on website
- ☒ Other publication
- ☐ Submission to regulatory authorities
- ☐ Access to raw data and right to publish freely by all investigators in study or by Independent Steering Committee on behalf of all investigators
- ☐ No plans to report or disseminate the results
- ☐ Other (please specify)

**A53. Will you inform participants of the results?**

☒ Yes ☐ No

Please give details of how you will inform participants or justify if not doing so.

Participants will not be informed about their individual survey results as they will be pooled together with those of other participants for analysis.

The research team will be making regular updates on the project website and Facebook and Twitter pages. Participants can be kept up-to-date with the research by accessing these pages. For participants who have indicated their willingness to receive regular electronic updates, we will send them regular e-newsletters about the results and the findings from similar research.

**5. Scientific and Statistical Review**

**A54. How has the scientific quality of the research been assessed? Tick as appropriate:**

- ☐ Independent external review
- ☐ Review within a company
- ☐ Review within a multi-centre research group
- ☐ Review within the Chief Investigator's institution or host organisation
- ☐ Review within the research team
- ☒ Review by educational supervisor
- ☐ Other

Justify and describe the review process and outcome. If the review has been undertaken but not seen by the researcher, give details of the body which has undertaken the review:

The proposal for this research study was reviewed initially by the internal research team and Trustees of the funding charity Cerebra as a part of the process to decide whether to award funding to Professor Hastings as a Cerebra Academic Chair. The detailed protocol was reviewed and developed by Professor Hastings and Dr Vaso Totsika as supervisors of three PhD students who will be responsible for working together to recruit to this study and to analyse key data.

For all studies except non-doctoral student research, please enclose a copy of any available scientific critique reports, together with any related correspondence.

For non-doctoral student research, please enclose a copy of the assessment from your educational supervisor/ institution.

**A56. How have the statistical aspects of the research been reviewed?** *Tick as appropriate:*

- ☐ Review by independent statistician commissioned by funder or sponsor  
☐ Other review by independent statistician  
☐ Review by company statistician  
☐ Review by a statistician within the Chief Investigator's institution  
☐ Review by a statistician within the research team or multi-centre group  
☒ Review by educational supervisor  
☐ Other review by individual with relevant statistical expertise  
☐ No review necessary as only frequencies and associations will be assessed – details of statistical input not required

*In all cases please give details below of the individual responsible for reviewing the statistical aspects. If advice has been provided in confidence, give details of the department and institution concerned.*

	Title Forename/Initials Surname
	Dr Vaso Totsika
Department	Centre for Educational Development, Appraisal and Research (CEDAR)
Institution	University of Warwick
Work Address	Coventry
Post Code	CV47AL
Telephone	024 765 22185
Fax	
Mobile	
E-mail	V.Totsika@warwick.ac.uk

*Please enclose a copy of any available comments or reports from a statistician.*

**A57. What is the primary outcome measure for the study?**

The primary outcome for this study depends on which of several different research aims is being addressed. For some analyses, the outcomes of interest will be parental well-being (e.g., stress, mental health, positive perceptions), and for others the outcomes will be child behavioural and emotional problems or adaptive skills.

**A58. What are the secondary outcome measures?** *(if any)***A59. What is the sample size for the research?** *How many participants/samples/data records do you plan to study in total? If there is more than one group, please give further details below.*

Total UK sample size: 4000  
 Total international sample size (including UK): 4000  
 Total in European Economic Area: 0

*Further details:*

Our aim is to recruit at least 1000 (and up to 2000) families of children with an intellectual disability. If two parental caregivers responded from 2000 families, the maximum number of participants would be 4000.

**A60. How was the sample size decided upon?** *If a formal sample size calculation was used, indicate how this was done,*

*giving sufficient information to justify and reproduce the calculation.*

The target number of families is set to at least 1000, in part because the aim is for this to be the largest survey on families of a child with an intellectual disability in the UK. Importantly, a large sample size in Wave 1 is imperative to ensure a good sample size in Wave 2, to allow for attrition.

To examine the power of the proposed (minimum) sample size of 1,000 families, we need to consider (a) the nature of the data, and (b) the most analytically complex and demanding models that will be employed to address the research aims.

All data collected will come from families and will refer to at least two family members (e.g., mothers-fathers, mother-child; mother-sibling). Any type of modelling will need to account for the clustering of participants within families. At the micro-level (Level 1) we have data for each individual family member (child, mother, father, sibling). At the macro-level (Level 2) we have information about the family as a whole (e.g., family income). Individuals who cluster within the same level (i.e., family) are expected to have outcomes more similar than individuals from different clusters. We, therefore, need to account for this potential association in all our models.

There are two multivariate approaches that are appropriate for this type of data: (a) multi-level models (also known as mixed models), and (b) generalised estimating equations. Both will be used to address research questions in this project. They are appropriate for analysing clustered data, and are a type of hierarchical regression analysis. These models will explore associations between child, mother, father, sibling, and overall family environment factors (e.g., family economic position) associated with child outcomes (e.g., child behaviour problems). Standard regression models cannot be fitted to this type of data as the hierarchical nature of the data violates the assumption of independence (as noted above), whereas multi-level models not only accommodate this but treat it as a substantive part of the analysis. Generalised estimating equations can accommodate the issue of clustering by accounting for its effect on the substantive part of the modelling. Power analyses for generalised estimating equations in family research have not been well developed yet, but the rationale and guidance will be similar to that for multi-level models, with one caveat. Available information on linear multilevel models indicates that sample size is equally important at every level of the investigation, and for this we will aim to achieve a large sample of participating families and children (i.e., sample sufficient numbers at the micro and macro level). Available guidance for generalised estimating equations (Garson, 2013) indicates that sample size is more important for level 2 (i.e., the macro-level) variables. For the present design (one child with intellectual disability recruited per family), this suggests that Level 2 and Level 1 sample sizes should be targeted at the same rate (i.e., if we need 1,000 children we need to target 1,000 families, and vice versa).

Available guidance on linear 2-level multilevel models suggests that power of about .80 for level 1 fixed effect predictors can be achieved by samples of between 600 to 1200 level-1 participants (Bell et al., 2010). Assuming more demanding models with 20 covariates, and low levels of intra-class correlation (0.10; a reasonable expectation on the basis of our previous research; Jones et al., 2013), 1,000 level 1 participants would allow power of about 1.0 to detect a small effect size (.30) in a 2-level design (Hedges & Rhoads, 2010). Therefore, the power analysis suggests that a proposed sample size of 1,000 level 1 participants is powerful enough to detect at least a small effect size in presence of several model covariates and low intra-class correlation. In this way, power for analytically less demanding models will also be sufficient.

**A61. Will participants be allocated to groups at random?**

☐ Yes ☒ No

**A62. Please describe the methods of analysis (statistical or other appropriate methods, e.g. for qualitative research) by which the data will be evaluated to meet the study objectives.**

SPSS software will be used to perform statistical analysis according to the research aims.

Levels of well-being in mothers, fathers and siblings will be analysed through descriptive statistics and one-sample t-tests (and non-parametric alternatives where appropriate). This will be compared to levels of well-being in families from normative data and other UK samples.

Multi-level modelling will be used to explore well-being scores, relationship quality, and parenting attitudes and beliefs between mothers and fathers in the same families.

Factors associated with the well-being of mothers, fathers and siblings will be explored through correlation and regression approaches. This will also be used to explore the factors associated with the well-being of the child with the intellectual disability. Final models will be tested using structural equation modelling.

Structural equation modelling approaches will be used for the longitudinal analyses to focus on factors associated with the well-being of the child with the intellectual disability and family members over time.

## 6. MANAGEMENT OF THE RESEARCH

**A63. Other key investigators/collaborators.** Please include all grant co-applicants, protocol co-authors and other key members of the Chief Investigator's team, including non-doctoral student researchers.

	Title	Forename/Initials	Surname
	Ms	Elizabeth	Halstead

Post Visiting Fellow

Qualifications BSc Psychology

Employer Cerebra

Work Address CEDAR  
University of Warwick

Post Code CV4 7AL

Telephone

Fax

Mobile

Work Email E.Halstead@warwick.ac.uk

	Title	Forename/Initials	Surname

Post

Qualifications

Employer

Work Address

Post Code

Telephone

Fax

Mobile

Work Email

	Title	Forename/Initials	Surname

Post

Qualifications

Employer

Work Address

Post Code

Telephone

Fax

Mobile

NHS REC Form

Reference:  
15/WM/0267

IRAS Version 4.0.0

Work Email
Title Forename/Initials Surname
Post
Qualifications
Employer
Work Address
Post Code
Telephone
Fax
Mobile
Work Email

## A64. Details of research sponsor(s)

## A64-1. Sponsor

## Lead Sponsor

- Status: ☐ NHS or HSC care organisation  
☒ Academic  
☐ Pharmaceutical industry  
☐ Medical device industry  
☐ Local Authority  
☐ Other social care provider (including voluntary sector or private organisation)  
☐ Other

Commercial status: Non-Commercial

*If Other, please specify:*

## Contact person

Name of organisation University of Warwick  
 Given name Jane  
 Family name Prewett  
 Address Research and Impact Services  
 Town/city Coventry  
 Post code CV4 8UW  
 Country UNITED KINGDOM  
 Telephone 02476522746  
 Fax  
 E-mail WMSSponsorship@warwick.ac.uk

## Is the sponsor based outside the UK?

☐ Yes ☒ No

*Under the Research Governance Framework for Health and Social Care, a sponsor outside the UK must appoint a*

legal representative established in the UK. Please consult the guidance notes.

**A65. Has external funding for the research been secured?**

- ☒ Funding secured from one or more funders  
☐ External funding application to one or more funders in progress  
☐ No application for external funding will be made

What type of research project is this?

- ☒ Standalone project  
☐ Project that is part of a programme grant  
☐ Project that is part of a Centre grant  
☐ Project that is part of a fellowship/ personal award/ research training award  
☐ Other

Other – please state:

Please give details of funding applications.

Organisation Cerebra  
 Address 2nd Floor Offices, The Lyric Buildings  
 King Street  
 Carmarthen  
 Post Code SA31 1BD  
 Telephone 01267244200  
 Fax  
 Mobile  
 Email TracyE@cerebra.org.uk

Funding Application Status: ☒ Secured ☐ In progress

Amount: 270,000

Duration  
 Years: 6  
 Months:

If applicable, please specify the programme/ funding stream:

What is the funding stream/ programme for this research project?

**A67. Has this or a similar application been previously rejected by a Research Ethics Committee in the UK or another country?**

- ☐ Yes ☒ No

Please provide a copy of the unfavourable opinion letter(s). You should explain in your answer to question A6-2 how the reasons for the unfavourable opinion have been addressed in this application.

NHS REC Form

Reference:  
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**A69-1. How long do you expect the study to last in the UK?**

Planned start date: 01/09/2015

Planned end date: 31/12/2019

Total duration:

Years: 4 Months: 3 Days: 31

**A71-2. Where will the research take place? (Tick as appropriate)**

- ☒ England  
☒ Scotland  
☒ Wales  
☒ Northern Ireland  
☐ Other countries in European Economic Area

Total UK sites in study 1

Does this trial involve countries outside the EU?

☐ Yes ☒ No
**A72. What host organisations (NHS or other) in the UK will be responsible for the research sites? Please indicate the type of organisation by ticking the box and give approximate numbers of planned research sites:**

- ☐ NHS organisations in England  
☐ NHS organisations in Wales  
☐ NHS organisations in Scotland  
☐ HSC organisations in Northern Ireland  
☐ GP practices in England  
☐ GP practices in Wales  
☐ GP practices in Scotland  
☐ GP practices in Northern Ireland  
☐ Social care organisations  
☐ Phase 1 trial units  
☐ Prison establishments  
☐ Probation areas  
☐ Independent hospitals  
☒ Educational establishments 1  
☐ Independent research units  
☐ Other (give details)

Total UK sites in study: 1

**A76. Insurance/ indemnity to meet potential legal liabilities**

*Note: in this question to NHS indemnity schemes include equivalent schemes provided by Health and Social Care (HSC) in Northern Ireland*

**A76-1. What arrangements will be made for insurance and/or indemnity to meet the potential legal liability of the sponsor(s) for harm to participants arising from the management of the research? Please tick box(es) as applicable.**

*Note: Where a NHS organisation has agreed to act as sponsor or co-sponsor, indemnity is provided through NHS schemes. Indicate if this applies (there is no need to provide documentary evidence). For all other sponsors, please describe the arrangements and provide evidence.*

- ☐ NHS indemnity scheme will apply (NHS sponsors only)
- ☒ Other insurance or indemnity arrangements will apply (give details below)

The University of Warwick has in force a Public and Products liability policy, a Clinical Trials insurance policy and a Professional Indemnity policy which provides cover for "negligent harm" and the activities here are included with in that coverage subject to the terms, conditions and exceptions of the policy.

*Please enclose a copy of relevant documents.*

**A76-2. What arrangements will be made for insurance and/ or indemnity to meet the potential legal liability of the sponsor(s) or employer(s) for harm to participants arising from the design of the research? Please tick box(es) as applicable.**

*Note: Where researchers with substantive NHS employment contracts have designed the research, indemnity is provided through NHS schemes. Indicate if this applies (there is no need to provide documentary evidence). For other protocol authors (e.g. company employees, university members), please describe the arrangements and provide evidence.*

- ☐ NHS indemnity scheme will apply (protocol authors with NHS contracts only)
- ☒ Other insurance or indemnity arrangements will apply (give details below)

The University of Warwick has in force a Public and Products liability policy, a Clinical Trials insurance policy and a Professional Indemnity policy which provides cover for "negligent harm" and the activities here are included with in that coverage subject to the terms, conditions and exceptions of the policy.

*Please enclose a copy of relevant documents.*

**A76-3. What arrangements will be made for insurance and/ or indemnity to meet the potential legal liability of investigators/collaborators arising from harm to participants in the conduct of the research?**

*Note: Where the participants are NHS patients, indemnity is provided through the NHS schemes or through professional indemnity. Indicate if this applies to the whole study (there is no need to provide documentary evidence). Where non-NHS sites are to be included in the research, including private practices, please describe the arrangements which will be made at these sites and provide evidence.*

- ☐ NHS indemnity scheme or professional indemnity will apply (participants recruited at NHS sites only)
- ☒ Research includes non-NHS sites (give details of insurance/ indemnity arrangements for these sites below)

The University of Warwick has in force a Public and Products liability policy, a Clinical Trials insurance policy and a Professional Indemnity policy which provides cover for "negligent harm" and the activities here are included with in that coverage subject to the terms, conditions and exceptions of the policy.

*Please enclose a copy of relevant documents.*



**PART C: Overview of research sites**

**Please enter details of the host organisations (Local Authority, NHS or other) in the UK that will be responsible for the research sites.** For NHS sites, the host organisation is the Trust or Health Board. Where the research site is a primary care site, e.g. GP practice, please insert the host organisation (PCT or Health Board) in the Institution row and insert the research site (e.g. GP practice) in the Department row.

Research site		Investigator/ Collaborator/ Contact	
Institution name	University of Warwick	Title	Professor
Department name	Centre for Educational Development Appraisal and Research	First name/ Initials	Richard
Street address	University of Warwick	Surname	Hastings
Town/city	Coventry		
Post Code	CV4 7AL		

**PART D: Declarations****D1. Declaration by Chief Investigator**

1. The information in this form is accurate to the best of my knowledge and belief and I take full responsibility for it.
2. I undertake to abide by the ethical principles underlying the Declaration of Helsinki and good practice guidelines on the proper conduct of research.
3. If the research is approved I undertake to adhere to the study protocol, the terms of the full application as approved and any conditions set out by review bodies in giving approval.
4. I undertake to notify review bodies of substantial amendments to the protocol or the terms of the approved application, and to seek a favourable opinion from the main REC before implementing the amendment.
5. I undertake to submit annual progress reports setting out the progress of the research, as required by review bodies.
6. I am aware of my responsibility to be up to date and comply with the requirements of the law and relevant guidelines relating to security and confidentiality of patient or other personal data, including the need to register when necessary with the appropriate Data Protection Officer. I understand that I am not permitted to disclose identifiable data to third parties unless the disclosure has the consent of the data subject or, in the case of patient data in England and Wales, the disclosure is covered by the terms of an approval under Section 251 of the NHS Act 2006.
7. I understand that research records/data may be subject to inspection by review bodies for audit purposes if required.
8. I understand that any personal data in this application will be held by review bodies and their operational managers and that this will be managed according to the principles established in the Data Protection Act 1998.
9. I understand that the information contained in this application, any supporting documentation and all correspondence with review bodies or their operational managers relating to the application:
  - Will be held by the REC (where applicable) until at least 3 years after the end of the study; and by NHS R&D offices (where the research requires NHS management permission) in accordance with the NHS Code of Practice on Records Management.
  - May be disclosed to the operational managers of review bodies, or the appointing authority for the REC (where applicable), in order to check that the application has been processed correctly or to investigate any complaint.
  - May be seen by auditors appointed to undertake accreditation of RECs (where applicable).
  - Will be subject to the provisions of the Freedom of Information Acts and may be disclosed in response to requests made under the Acts except where statutory exemptions apply.
  - May be sent by email to REC members.
10. I understand that information relating to this research, including the contact details on this application, may be held on national research information systems, and that this will be managed according to the principles established in the Data Protection Act 1998.
11. Where the research is reviewed by a REC within the UK Health Departments Research Ethics Service, I understand that the summary of this study will be published on the website of the National Research Ethics Service (NRES), together with the contact point for enquiries named below. Publication will take place no earlier than 3 months after issue of the ethics committee's final opinion or the withdrawal of the application.

**Contact point for publication(Not applicable for R&D Forms)**

NRES would like to include a contact point with the published summary of the study for those wishing to seek further information. We would be grateful if you would indicate one of the contact points below.

- ☒ Chief Investigator  
☐ Sponsor

NHS REC Form

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- ☐ Study co-ordinator  
☐ Student  
☐ Other – please give details  
☐ None

**Access to application for training purposes** *(Not applicable for R&D Forms)**Optional – please tick as appropriate:*

☒ I would be content for members of other RECs to have access to the information in the application in confidence for training purposes. All personal identifiers and references to sponsors, funders and research units would be removed.

This section was signed electronically by Professor Richard Hastings on 13/07/2015 15:03.

Job Title/Post: Professor  
Organisation: University of Warwick  
Email: R.Hastings@warwick.ac.uk

**D2. Declaration by the sponsor's representative**

*If there is more than one sponsor, this declaration should be signed on behalf of the co-sponsors by a representative of the lead sponsor named at A64-1.*

I confirm that:

1. This research proposal has been discussed with the Chief Investigator and agreement in principle to sponsor the research is in place.
2. An appropriate process of scientific critique has demonstrated that this research proposal is worthwhile and of high scientific quality.
3. Any necessary indemnity or insurance arrangements, as described in question A76, will be in place before this research starts. Insurance or indemnity policies will be renewed for the duration of the study where necessary.
4. Arrangements will be in place before the study starts for the research team to access resources and support to deliver the research as proposed.
5. Arrangements to allocate responsibilities for the management, monitoring and reporting of the research will be in place before the research starts.

*Please note: The declarations below do not form part of the application for approval above. They will not be considered by the Research Ethics Committee.*

6. Where the research is reviewed by a REC within the UK Health Departments Research Ethics Service, I understand that the summary of this study will be published on the website of the National Research Ethics Service (NRES), together with the contact point for enquiries named in this application. Publication will take place no earlier than 3 months after issue of the ethics committee's final opinion or the withdrawal of the application.
7. Specifically, for submissions to the Research Ethics Committees (RECs) I declare that any and all clinical trials approved by the HRA since 30th September 2013 (as defined on IRAS categories as clinical trials of medicines, devices, combination of medicines and devices or other clinical trials) have been registered on a publically accessible register in compliance with the HRA registration requirements for the UK, or that any deferral granted by the HRA still applies.

This section was signed electronically by Mrs Jane Prewett on 13/07/2015 23:33.

Job Title/Post: Deputy Director, R&IS  
 Organisation: University of Warwick  
 Email: jane.prewett@warwick.ac.uk

**D3. Declaration for student projects by academic supervisor(s)**

1. I have read and approved both the research proposal and this application. I am satisfied that the scientific content of the research is satisfactory for an educational qualification at this level.
2. I undertake to fulfil the responsibilities of the supervisor for this study as set out in the Research Governance Framework for Health and Social Care.
3. I take responsibility for ensuring that this study is conducted in accordance with the ethical principles underlying the Declaration of Helsinki and good practice guidelines on the proper conduct of research, in conjunction with clinical supervisors as appropriate.
4. I take responsibility for ensuring that the applicant is up to date and complies with the requirements of the law and relevant guidelines relating to security and confidentiality of patient and other personal data, in conjunction with clinical supervisors as appropriate.

**Academic supervisor 1**

This section was signed electronically by Dr Vasiliki Totsika on 13/07/2015 21:33.

Job Title/Post: Senior Research Fellow  
Organisation: University of Warwick  
Email: V.Totsika@warwick.ac.uk

**Academic supervisor 2**

This section was signed electronically by Professor Richard Hastings on 13/07/2015 15:04.

Job Title/Post: Professor  
Organisation: University of Warwick  
Email: R.Hastings@warwick.ac.uk

## Appendix C: Information sheet (Primary Caregiver) pre age amendment



# 1,000 Families study

We would like to invite you to take part in our research study. Before you decide to take part it is important for you to understand what the research would involve. Please take time to read the following information carefully. If there is anything that is unclear, or if you would like more information please contact us using the details provided at the end.

### Purpose of the study

The aim of this study is to understand the experiences of family members living in the UK who care for a child with an intellectual disability (often called learning disability or learning difficulties in the UK). We wish to capture the experiences of parents/parental caregivers and any siblings. We will be recruiting a large number of families (at least 1,000 Families) to take part in this research. We would also like to follow families over time, because it is important to understand how families change over time and adjust in different ways to having a child with intellectual disability.

### Who can take part?

We are inviting primary parental caregivers of children with intellectual disability (learning disability or learning difficulties) between the ages of 4 and 11 years to take part in this research. Primary parental caregivers might not be the child's mother, but would be the adult who cares for the child with intellectual disability for most of the time. Mothers may be biological, adoptive, or foster mothers. Your child with intellectual disability might also have other diagnosed conditions such as autism, Down syndrome, or other genetic syndromes. Your family must

currently live somewhere in the UK. This research focuses on families whose child with intellectual disability lives with them for the majority of the time (more than half of a typical week). If your child with intellectual disability lives outside of the family home for the majority of the week (e.g., in a residential school placement) then this research study is not for you.

### **Why have I been invited to take part in the research?**

You have been invited to take part in this research because you are a parent or parental caregiver of a child with intellectual disability aged between 4 and 11 years and living in the UK.

### **What will happen if I take part?**

Once you have read this study information, you will be asked to read some statements and to indicate your agreement to each one. We need to check that you agree to participate in the research. The online survey then includes questions asking about you and your family, your experiences as a parent, about your child with intellectual disability, and also about a sibling in the family if there is one. We expect the online survey to take about 20 minutes to complete in total.

If you agree, a researcher will then telephone you to ask you some additional questions about your child with intellectual disability. These questions are much easier to ask in a telephone interview, and focus on understanding the skills that your child with intellectual disability has and on some of the difficulties that they may face. This telephone interview usually takes about 40-60 minutes. We can schedule the interview for a time that suits you, and it is easy to re-arrange a time if the first time organised ends up being inconvenient for you.

If you agree, we will send you updates about this research project and other research that we are carrying out about the experiences of families of individuals with intellectual disability.

We plan to follow up with 1,000 Families Study families in about two years' time. If you agree, we will contact you again then to ask if you would like to take part in this follow-up research. At that stage, we would ask you to complete the online survey and the telephone interview again.

### **Do I have to take part in the research?**

You are under no obligation to participate in the research, your involvement is voluntary. You can decide to complete the online survey and not the telephone interview. You can also complete the online survey and telephone interview, but not give us permission to invite you to take part again in two years' time.

If you do agree to participate and then decide you no longer want to take part, you are free to withdraw. You do not have to provide a reason for wanting to withdraw from the study. You would have the option for any data you have provided up until your withdrawal to be removed from the study and destroyed.

You are able to withdraw yourself from the study even if another parent/caregiver from your household has also participated in the study.

Apart from the questions checking that you are happy to take part in the various parts of the study, you do not have to answer any questions that you do not wish to answer.

### **Are there any possible benefits and risks of taking part in the study?**

Involvement in this research provides an opportunity to share your and your family's experiences of raising a child with an intellectual disability. The information you provide will help us to understand more about families like yours, to share this information widely, and to inform ways to better support families.

We do not anticipate any risks to taking part in this research. The questions that we are asking have been used in several research studies before, and we have removed any that have tended to cause family members distress. However, it is possible that you will find some of the questions to be upsetting because we do ask about your well-being and some of the difficulties faced by you, your family and your child with intellectual disability. We do also ask about positive experiences. If you are upset by any of the questions, you do not have to respond to them and you are under no obligation to continue with the survey or interview.

If any of the survey or interview questions make you concerned for yourself or another family member's well-being, we recommend that you make contact with your General Practitioner (GP) or one of the helplines listed below:

Mencap: 0808 808 1111, Contact a Family: 01332 557 975, KIDS: 0207 359 3635

### **How will the information you give to us be looked after?**

All information that you provide as a part of this study will remain confidential, and we will store the information securely (in locked cabinets, or secure password protected computers) in an anonymised form. Access is restricted to the research team.



We will publish reports and give presentations about the results of the study. However, you will not be identified individually in any way as your responses will be pooled together with other participants and you will be assigned an anonymised study number. You and your family will not be able to be identified in any report or presentation about the study.

There are circumstances in which we would not be able to keep confidential something that you say. If you mention during the telephone interview any information that suggests someone in your family, or you, is at risk of harm or has been subject to abuse, the researcher would have a duty to report this information to the appropriate authorities.

We will keep your personal data for three years so that we can invite you to take part in the research study again in approximately two years' time. At that time, we will check again if we can approach you again in the future. Your anonymised responses to the survey and interview questions will be archived so that researchers in the future can carry out additional analysis of the data from the 1,000 Families Study. These researchers would not have access to your personal information and would have to agree to abide by appropriate ethical principles to do any new research.

## **Who is organising and funding the research?**

The University of Warwick is responsible for this research. The research has received funding from the charity Cerebra and through the Economic and Social Research Council Doctoral Training Centre at the University of Warwick.

## **Who has reviewed the study?**

This study has been reviewed and given a favourable opinion by a NHS Research Ethics Committee. A Research Ethics Committee is a group of independent people who review research to protect the dignity, rights, safety, and well-being of participants and researchers.

## **Further information and contact details**

If you would like to ask questions before deciding whether to participate, please contact a member of the research team (Tel: 02476 524 139, Email: [familyresearch@warwick.ac.uk](mailto:familyresearch@warwick.ac.uk)).

If you are unhappy about any aspect of this research study, please contact the Chief Investigator Richard Hastings by mail, email or telephone (CEDAR, University of Warwick, Coventry, CV4 7AL; [R.Hastings@warwick.ac.uk](mailto:R.Hastings@warwick.ac.uk); 02476 522 197).

*If you would like to discuss the research, or ask any questions, with **someone who is not a part of the study team**, please contact Ms.Charlotte Rowe (Email: C.Rowe.1@warwick.ac.uk; 02476 524 139).*

This study is covered by the University of Warwick's insurance and indemnity cover.

Any complaint about the way that you have been dealt with during the study, or any possible harm that you might have suffered, should be directed to: the Director of Delivery Assurance, Registrar's Office, University House, University of Warwick, Coventry, CV4 8UW. email – complaints@warwick.ac.uk; telephone 02476 574 774

**If you are happy to be involved in the study, please go  
to  
[www.surveymonkey.com/r/1000families](http://www.surveymonkey.com/r/1000families)**

**Appendix D: Consent form (Primary Caregiver)**

# 1,000 Families study

## Primary caregiver consent form

*Please read carefully the initial statements below. If you agree with these statements then tick in the corresponding box.*

- ☐ I confirm that I have read and understood the information provided for the 1,000 Families study. I have had the opportunity to consider the information, ask questions and have had these answered to my satisfaction.
- ☐ I understand that my participation is voluntary and that I am free to withdraw at any time, without giving any reason, and without my rights being affected.
- ☐ I understand that relevant sections of my data collected during the study, may be looked at by individuals from regulatory authorities responsible for investigating research fraud. I give permission for individuals from these regulatory authorities to have access to my records if research fraud in this study was to be investigated.
- ☐ I agree to participate in the survey.

**Appendix E: Online survey (Primary and Secondary Caregiver)**

Thank you for taking part in the 1,000 Families Study. Your participation in this survey will make a valuable contribution to UK based family and disability research.

We estimate that the survey will take around 20 minutes for you to complete.

On the next page you will be asked to specify whether you are the primary or secondary caregiver to your child with intellectual disability and will be directed to in-depth information about the study as required and approved by the NHS Ethics Committee. You will have the option to download a PDF copy of this information to read at a time of your convenience.

Please click 'Next' to start the survey.

## Online survey

1. Are you the primary or secondary caregiver to your child with intellectual disability?

- ☐ Primary Caregiver
- ☐ Secondary Caregiver

## Primary Caregiver Information

[Download and keep a PDF version of the information sheet here](#)

The PDF download includes exactly the same information as you can read below.

We would like to invite you to take part in our research study. Before you decide to take part it is important for you to understand what the research would involve. Please take time to read the following information carefully. If there is anything that is unclear, or if you would like more information please contact us using the details provided at the end.

### **Purpose of the study**

The aim of this study is to understand the experiences of family members living in the UK who care for a child with an intellectual disability (often called learning disability or learning difficulties in the UK). We wish to capture the experiences of parents/parental caregivers and any siblings. We will be recruiting a large number of families (at least 1,000 Families) to take part in this research. We would also like to follow families over time, because it is important to understand how families change over time and adjust in different ways to having a child with intellectual disability.

### **Who can take part?**

We are inviting primary parental caregivers of children with intellectual disability (learning disability or learning difficulties) between the ages of 4 and 11 years to take part in this research. Primary parental caregivers might not be the child's mother, but would be the adult who cares for the child with intellectual disability for most of the time. Mothers may be biological, adoptive, or foster mothers. Your child with intellectual disability might also have other diagnosed conditions such as autism, Down syndrome, or other genetic syndromes. Your family must currently live somewhere in the UK. This research focuses on families whose child with intellectual disability lives with them for the majority of the time (more than half of a typical week). If your child with intellectual disability lives outside of the family home for the majority of the week (e.g., in a residential school placement) then this research study is not for you.

### **Why have I been invited to take part in the research?**

You have been invited to take part in this research because you are a parent or parental caregiver of a child with intellectual disability aged between 4 and 11 years and living in the UK.

### **What will happen if I take part?**

Once you have read this study information, you will be asked to read some statements and to indicate your agreement to each one. We need to check that you agree to participate in the research. The online survey then includes questions asking about you and your family, your experiences as a parent, about your child with intellectual disability, and also about a sibling in the family if there is one. We expect the online survey to take about 20 minutes to complete in total.

If you agree, a researcher will then telephone you to ask you some additional questions about your child with intellectual disability. These questions are much easier to ask in a telephone interview, and focus on understanding the skills that your child with intellectual disability has and on some of the difficulties that they may face. This telephone interview usually takes about 40-60 minutes. We can schedule the interview for a time that suits you, and it is easy to re-arrange a time if the first time organised ends up being inconvenient for you.

If you agree, we will send you updates about this research project and other research that we are carrying out about the experiences of families of individuals with intellectual disability.

We plan to follow up with 1,000 Families Study families in about two years' time. If you agree, we will contact you again then to ask if you would like to take part in this follow-up research. At that stage, we would ask you to complete the online survey and the telephone interview again.

**Do I have to take part in the research?**

You are under no obligation to participate in the research, your involvement is voluntary. You can decide to complete the online survey and not the telephone interview. You can also complete the online survey and telephone interview, but not give us permission to invite you to take part again in two years' time.

If you do agree to participate and then decide you no longer want to take part, you are free to withdraw. You do not have to provide a reason for wanting to withdraw from the study. You would have the option for any data you have provided up until your withdrawal to be removed from the study and destroyed.

You are able to withdraw yourself from the study even if another parent/caregiver from your household has also participated in the study.

Apart from the questions checking that you are happy to take part in the various parts of the study, you do not have to answer any questions that you do not wish to answer.

**Are there any possible benefits and risks of taking part in the study?**

Involvement in this research provides an opportunity to share your and your family's experiences of raising a child with an intellectual disability. The information you provide will help us to understand more about families like yours, to share this information widely, and to inform ways to better support families.

We do not anticipate any risks to taking part in this research. The questions that we are asking have been used in several research studies before, and we have removed any that have tended to cause family members distress. However, it is possible that you will find some of the questions to be upsetting because we do ask about your well-being and some of the difficulties faced by you, your family and your child with intellectual disability. We do also ask about positive experiences. If you are upset by any of the questions, you do not have to respond to them and you are under no obligation to continue with the survey or interview.

If any of the survey or interview questions make you concerned for yourself or another family member's well-being, we recommend that you make contact with your General Practitioner (GP) or one of the helplines listed below:

Mencap: 0808 808 1111, Contact a Family: 01332 557 975, KIDS: 0207 359 3635



**How will the information you give to us be looked after?**

All information that you provide as a part of this study will remain confidential, and we will store the information securely (in locked cabinets, or secure password protected computers) in an anonymised form. Access is restricted to the research team.

We will publish reports and give presentations about the results of the study. However, you will not be identified individually in any way as your responses will be pooled together with other participants and you will be assigned an anonymised study number. You and your family will not be able to be identified in any report or presentation about the study.

There are circumstances in which we would not be able to keep confidential something that you say. If you mention during the telephone interview any information that suggests someone in your family, or you, is at risk of harm or has been subject to abuse, the researcher would have a duty to report this information to the appropriate authorities.

We will keep your personal data for three years so that we can invite you to take part in the research study again in approximately two years' time. At that time, we will check again if we can approach you again in the future. Your anonymised responses to the survey and interview questions will be archived so that researchers in the future can carry out additional analysis of the data from the 1,000 Families Study. These researchers would not have access to your personal information and would have to agree to abide by appropriate ethical principles to do any new research.

**Who is organising and funding the research?**

The University of Warwick is responsible for this research. The research has received funding from the charity Cerebra and through the Economic and Social Research Council Doctoral Training Centre at the University of Warwick.

**Who has reviewed the study?**

This study has been reviewed and given a favourable opinion by a NHS Research Ethics Committee. A Research Ethics Committee is a group of independent people who review research to protect the dignity, rights, safety, and well-being of participants and researchers.

**Further information and contact details**

If you would like to ask questions before deciding whether to participate, please contact a member of the research team (Tel: 02476 524 139, Email: [familyresearch@warwick.ac.uk](mailto:familyresearch@warwick.ac.uk)).

If you are unhappy about any aspect of this research study, please contact the Chief Investigator Richard Hastings by mail, email or telephone (CEDAR, University of Warwick, Coventry, CV4 7AL; [R.Hastings@warwick.ac.uk](mailto:R.Hastings@warwick.ac.uk); 02476 522 197).

*If you would like to discuss the research, or ask any questions, with **someone who is not a part of the study team**, please contact Ms. Charlotte Rowe (Email: [C.Rowe.1@warwick.ac.uk](mailto:C.Rowe.1@warwick.ac.uk); 02476 524 139).*

This study is covered by the University of Warwick's insurance and indemnity cover.

Any complaint about the way that you have been dealt with during the study, or any possible harm that you might have suffered, should be directed to: the Director of Delivery Assurance, Registrar's Office, University House, University of Warwick, Coventry, CV4 8UW. email – [complaints@warwick.ac.uk](mailto:complaints@warwick.ac.uk); telephone 0247657 4774

**If you are happy to be involved in the study, please click 'Next' below to proceed to the consent questions.**

[Download and keep a PDF version of the information sheet here](#)

## Secondary Caregiver Information

[Download and keep a PDF version of the information sheet here](#)

The PDF download includes exactly the same information as you can read below.

We would like to invite you to take part in our research study. Before you decide to take part it is important for you to understand what the research would involve. Please take time to read the following information carefully. If there is anything that is unclear, or if you would like more information please contact us using the details provided at the end.

### **Purpose of the study**

The aim of this study is to understand the experiences of family members living in the UK who care for a child with an intellectual disability (often called learning disability or learning difficulties in the UK). We wish to capture the experiences of parents/parental caregivers and any siblings. We will be recruiting a large number of families (at least 1,000 Families) to take part in this research. We would also like to follow families over time, because it is important to understand how families change over time and adjust in different ways to having a child with intellectual disability.

### **Who can take part?**

We are inviting secondary parental caregivers of children with intellectual disability (learning disability or learning difficulties) between the ages of 4 and 11 years to take part in this research. Secondary parental caregivers might not be the child's father, but would be the adult who cares for the child with intellectual disability but not as much as the primary caregiver. The secondary caregiver might include a grandparent or the mother's partner. Fathers may be biological, adoptive, or step fathers. Your child with intellectual disability might also have other diagnosed conditions such as autism, Down syndrome, or other genetic syndromes. Your family must currently live somewhere in the UK. This research focuses on families whose child with intellectual disability lives with them for the majority of the time (more than half of a typical week). If your child with intellectual disability lives outside of the family home for the majority of the week (e.g., in a residential school placement) then this research study is not for you.

### **Why have I been invited to take part in the research?**

You have been invited to take part in this research because you are a parent or parental caregiver of a child with intellectual disability aged between 4 and 11 years and living in the UK.

### **What will happen if I take part?**

Once you have read this study information, you will be asked to read some statements and to indicate your agreement to each one. We need to check that you agree to participate in the research. The online survey then includes questions asking about you and your family, your experiences as the secondary caregiver, about your child with intellectual disability, and also about a sibling in the family if there is one. We expect the online survey to take about 20 minutes to complete in total.

If you agree, we will send you updates about this research project and other research that we are carrying out about the experiences of families of individuals with intellectual disability.

We plan to follow up with 1,000 Families Study families in about two years' time. If you agree, we will contact you again then to ask if you would like to take part in this follow-up research. At that stage, we would ask you to complete the online survey again.

**Do I have to take part in the research?**

You are under no obligation to participate in the research, your involvement is voluntary. You can complete the online survey but not give us permission to invite you to take part again in two years' time.

If you do agree to participate and then decide you no longer want to take part, you are free to withdraw. You do not have to provide a reason for wanting to withdraw from the study. You would have the option for any data you have provided up until your withdrawal to be removed from the study and destroyed.

You are able to withdraw yourself from the study even if another parent/caregiver from your household has also participated in the study.

Apart from the questions checking that you are happy to take part in the various parts of the study, you do not have to answer any questions that you do not wish to answer.

**Are there any possible benefits and risks of taking part in the study?**

Involvement in this research provides an opportunity to share you and your family's experiences of raising a child with an intellectual disability. The information you provide will help us to understand more about families like yours, to share this information widely, and to inform ways to better support families.

We do not anticipate any risks to taking part in this research. The questions that we are asking have been used in several research studies before, and we have removed any that have tended to cause family members distress. However, it is possible that you will find some of the questions to be upsetting because we do ask about your well-being and some of the difficulties faced by you, your family and your child with intellectual disability. We do also ask about positive experiences. If you are upset by any of the questions, you do not have to respond to them and you are under no obligation to continue with the survey.

If any of the survey questions make you concerned for yourself or another family member's well-being, we recommend that you make contact with your General Practitioner (GP) or one of the helplines listed below:

Mencap: 0808 808 1111, Contact a Family: 01332 557 975, KIDS: 0207 359 3635

**How will the information you give to us be looked after?**

All information that you provide as a part of this study will remain confidential, and we will store the information securely (in locked cabinets, or secure password protected computers) in an anonymised form. Access is restricted to the research team.

We will publish reports and give presentations about the results of the study. However, you will not be identified individually in any way as your responses will be pooled together with other participants and you will be assigned an anonymised study number. You and your family will not be able to be identified in any report or presentation about the study.

There are circumstances in which we would not be able to keep confidential something that you say. If you mention during the survey any information that suggests someone in your family, or you, is at risk of harm or has been subject to abuse, the researcher would have a duty to report this information to the appropriate authorities.

We will keep your personal data for three years so that we can invite you to take part in the research study again in approximately two years' time. At that time, we will check again if we can approach you again in the future. Your anonymised responses to the survey questions will be archived so that researchers in the future can carry out additional analysis of the data from the 1,000 Families Study. These researchers would not have access to your personal information and would have to agree to abide by appropriate ethical principles to do any new research.

**Who is organising and funding the research?**

The University of Warwick is responsible for this research. The research has received funding from the charity Cerebra and through the Economic and Social Research Council Doctoral Training Centre at the University of Warwick.

**Who has reviewed the study?**

This study has been reviewed and given a favourable opinion by a NHS Research Ethics Committee. A Research Ethics Committee is a group of independent people who review research to protect the dignity, rights, safety, and well-being of participants and researchers.

**Further information and contact details**

If you would like to ask questions before deciding whether to participate, please contact a member of the research team (Tel: 02476 524 139, Email: [familyresearch@warwick.ac.uk](mailto:familyresearch@warwick.ac.uk)).

If you are unhappy about any aspect of this research study, please contact the Chief Investigator Richard Hastings by mail, email or telephone (CEDAR, University of Warwick, Coventry, CV4 7AL; [R.Hastings@warwick.ac.uk](mailto:R.Hastings@warwick.ac.uk); 02476 522 197).

*If you would like to discuss the research, or ask any questions, with **someone who is not a part of the study team**, please contact Ms. Charlotte Rowe (Email: [C.Rowe.1@warwick.ac.uk](mailto:C.Rowe.1@warwick.ac.uk); 02476 524 139).*

This study is covered by the University of Warwick's insurance and indemnity cover.

Any complaint about the way that you have been dealt with during the study, or any possible harm that you might have suffered, should be directed to: the Director of Delivery Assurance, Registrar's Office, University House, University of Warwick, Coventry, CV4 8UW. email – [complaints@warwick.ac.uk](mailto:complaints@warwick.ac.uk); telephone 02476 574 774

**If you are happy to be involved in the study, please click 'Next' below to proceed to the consent questions.**

[Download and keep a PDF version of the information sheet here](#)

## Primary Caregiver Consent

***Please read carefully the initial statements below. If you agree with these statements then click the corresponding box.***

**2. If you agree with these statements then click the corresponding box.**

- ☐ I confirm that I have read and understood the Information provided for the 1,000 Families study. I have had the opportunity to consider the information, ask questions and have had these answered to my satisfaction.
- ☐ I understand that my participation is voluntary and that I am free to withdraw at any time, without giving any reason, and without my rights being affected.
- ☐ I understand that relevant sections of my data collected during the study, may be looked at by individuals from regulatory authorities responsible for investigating research fraud. I give permission for individuals from these regulatory authorities to have access to my records if research fraud in this study was to be investigated.
- ☐ I agree to participate in the online survey.

***Please enter the following details below so that your survey responses can be matched with any other respondents from your household.***

**3. Child with intellectual disability**

First name of your child  
with intellectual disability:

Surname of your child with  
intellectual disability:

**4. Your postcode**

Postcode

**5. If your child lives at a different address to you, please enter your child's postcode below**



6. I agree to participate in a telephone interview with a researcher (as described in the information sheet).

☐ Yes

☐ No

### Contacting you about the telephone interview

7. Please enter the following details so you can be contacted by a member of the research team to arrange a time for the telephone interview.

First name:

Surname:

#### 8. Telephone number

Home telephone number:

Mobile number:

#### 9. Email address

10. I agree that the research team can contact me in 2 years' time to invite me to participate in the follow up study.

☐ Yes

☐ No

### Contacting you in 2 years' time

11. Please provide the following details so that we can contact you for the follow-up study in 2 years' time.

First name:

Surname:

Address line 1:

Address line 2:

City/Town:

Postcode:

#### 12. Telephone number

Home telephone number:

Mobile number:

#### 13. Email address

### Email updates

14. I wish to be kept up to date by email with the progress and findings of the study and other research about families of individuals with intellectual disability.

- ☐ No
- ☐ Yes (please provide your email address)

## Secondary Caregiver Consent

***Please read carefully the initial statements below. If you agree with these statements then click the corresponding box.***

**15. If you agree with these statements then click the corresponding box.**

- ☐ I confirm that I have read and understood the Information provided for the 1,000 Families study. I have had the opportunity to consider the information, ask questions and have had these answered to my satisfaction.
- ☐ I understand that my participation is voluntary and that I am free to withdraw at any time, without giving any reason, and without my rights being affected.
- ☐ I understand that relevant sections of my data collected during the study, may be looked at by individuals from regulatory authorities responsible for investigating research fraud. I give permission for individuals from these regulatory authorities to have access to my records if research fraud in this study was to be investigated.
- ☐ I agree to participate in the online survey.

***Please enter the following details below so that your survey responses can be matched with any other respondents from your household.***

**16. Child with intellectual disability**

First name of your child  
with intellectual disability:

Surname of your child with  
intellectual disability:

**17. Your postcode**

Postcode

**18. If your child lives at a different address to you, please enter your child's postcode below**

19. I agree that the research team can contact me in 2 years' time to invite me to participate in the follow up study.

☐ Yes

☐ No

**Contacting you in 2 years' time**

20. Please provide the following details so that we can contact you for the follow-up study in 2 years' time.

First name:

Surname:

Address line 1:

Address line 2:

City/Town:

Postcode:

**21. Telephone number**

Home telephone number:

Mobile number:

**22. Email address**



### Email updates

23. I wish to be kept up to date by email with the progress and findings of the study and other research about families of individuals with intellectual disability.

- ☐ No
- ☐ Yes (please provide your email address)

### Questions about you and your child with Intellectual Disability

24. Is your child with intellectual disability male or female?

- ☐ Male
- ☐ Female

### *Questions about you*

25. Please indicate your relationship to the child with intellectual disability.

If Other please describe

26. How do you identify your gender?

- ☐ Male
- ☐ Female
- ☐ Trans
- ☐ Prefer not to answer

27. How would you describe your ethnicity?

28. If you wish to do so, please specify how you would describe your ethnicity. If you do not wish to provide more information, please ignore the question and click 'Next'.

29. Please select the highest level of your educational qualifications

30. Please select one option which best describes your status

- ☐ In a job and currently working for an employer
- ☐ On maternity/paternity /parental leave from a job
- ☐ Self-employed
- ☐ A Full time student
- ☐ Doing voluntary work
- ☐ Looking after home and family
- ☐ Unemployed
- ☐ Do something else (please specify below)

31. How is your health in general?

- ☐ Very good
- ☐ Good
- ☐ Fair
- ☐ Bad
- ☐ Very bad

32. Do you have a longstanding illness, disability or infirmity? By longstanding we mean anything that has troubled you over a period of time or is likely to affect you over a period of time?

- ☐ No
- ☐ Yes

### Questions about your household

33. In total how many people currently live in your home (including yourself)?

	Adults	Children
Number	<input type="text"/>	<input type="text"/>

*Data from research with families with a family member with a disability has shown that a family's financial resources are important in understanding family member's views and experiences. With this in mind, we would be grateful if you could answer the additional question below. We are not interested in exactly what your family income is, but we would like to be able to look at whether people with different levels of financial resources have different experiences.*

34. What is your total weekly household income (after any deductions e.g. income tax), including income from paid work, pension, Social Services Benefits (e.g. Job Seekers Allowance, DLA, Carers' Allowance, Attendance Allowance, Tax Credits, Housing Benefits, Pension Credits) etc.?

35. How well would you say you [and your husband/wife/partner] are managing financially these days?  
Would you say you are ...

- ☐ living comfortably?
- ☐ doing alright?
- ☐ just about getting by?
- ☐ finding it quite difficult?
- ☐ finding it very difficult?

36. Suppose you only had one week to raise £2000 for an emergency, which of the following best describes how hard it would be for you to get that money?

- ☐ I could easily raise the money
- ☐ I could raise the money, but it would involve some sacrifices (e.g. reduced spending, selling a possession)
- ☐ I would have to do something drastic to raise the money (e.g. selling an important possession)
- ☐ I don't think I could raise the money

## Questions about your child with intellectual disability

*We would now like to ask you some questions about your child with intellectual disability who you named earlier in the survey.*

37. The date of birth of your child with intellectual disability

	DD	MM	YYYY
Date	<input type="text"/>	<input type="text"/>	<input type="text"/>

38. Please select which of the conditions below professionals have diagnosed in relation to your child with intellectual disability (select all that apply)

- ☐ Learning disability/learning difficulty
- ☐ Autism/Autistic Spectrum Disorder /Autistic Spectrum Condition/Asperger's Syndrome
- ☐ Down syndrome
- ☐ Global Developmental delay
- ☐ Cerebral palsy
- ☐ Other genetic syndrome/diagnosis (please describe)

39. Please state if your child with intellectual disability has:

- ☐ A mild/moderate intellectual disability

Children with a mild to moderate intellectual disability can typically communicate and look after themselves well, but may take a bit longer to learn new skills compared to other children of the same age.

- ☐ A Severe/profound intellectual disability

Children with a severe to profound intellectual disability are likely to have complex and multiple difficulties which require extensive support to learn and carry out daily activities.

40. Does your child with intellectual disability have a visual impairment?

- ☐ Yes
- ☐ No

41. Does your child with intellectual disability have a hearing impairment?

☐ Yes

☐ No

42. Does your child with intellectual disability currently have epileptic seizures?

☐ Yes

☐ No

43. Has your child with intellectual disability ever had an epileptic seizure in the past?

☐ Yes

☐ No



44. Does your child with intellectual disability have any mobility problems?

☐ Yes

☐ No

45. Does your child with intellectual disability have any other physical health problems?

☐ Yes

☐ No

46. Does your child with intellectual disability normally

☐ Live with you full-time?

☐ Live with you part of the time?

47. Please state the approximate number of hours that your child lives with you on a weekly basis

48. What type of school does your child with intellectual disability usually attend?

- ☐ Mainstream school
- ☐ Mainstream school in either a special unit or resourced Special Educational Needs (SEN) provision
- ☐ Special school
- ☐ Home schooled
- ☐ Not currently in school

## Questions about your experiences

*The following statements ask about your experiences of having a child with intellectual disability.*

49. Please respond to all questions by selecting the response which best describes how you feel about each statement.

	Strongly agree	Agree	Neither agree or disagree	Disagree	Strongly disagree
Since having this child I have grown as a person	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
Having this child has helped me learn new things/ skills	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
Raising this child helps put my life into perspective	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
Since having this child, my family has become closer to one another	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
Since having this child, my family has become more tolerant and accepting	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
Since having this child I have become more determined to face up to challenges	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
Since having this child I have a greater understanding of other people	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>

*We would now like to ask you about the time you have to do leisure or social activities*

50. Has the ability to spend time doing leisure or social activities been affected by the assistance you give to your child with intellectual disability in any of the ways described below? (For all responses, this should be as a result of the caring and not for other reasons). Please select any that apply.

- ☐ Unable to socialise or take part in social or leisure activities at all (due to caring responsibilities)
- ☐ Reduced time with spouse or partner
- ☐ Reduced time with other family members
- ☐ Reduced time with friends
- ☐ Difficulties making new friends
- ☐ Reduced time spent doing sport or physical activity
- ☐ Reduced time spent doing pastime or hobby

*The following questions ask about how you have been feeling during the past 30 days.*

51. For each question, please click the number that best describes how often you had this feeling.

During the past 30 days, about how often do you feel

	All of the time	Most of the time	Some of the time	A little of the time	None of the time
...nervous?	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
...hopeless?	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
...restless or fidgety?	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
...so depressed that nothing could cheer you up?	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
...that everything was an effort?	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
...worthless?	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>

*We are now going to ask you about your satisfaction with life.*

52. Here is a scale from 1-10 where '1' means that you are completely dissatisfied and '10' means that you are completely satisfied. All things considered, please could you enter the number which corresponds with how satisfied or dissatisfied you are about the way your life has turned out so far.

[illegible]

53. What is your current marital status?

- ☐ Married and living with spouse/civil partner
- ☐ Living with partner
- ☐ Divorced/Separated/Single/Widowed/Not currently living with partner



## Questions about your family

*We would now like to ask you about how satisfied you are with family life.*

56. Please read the following statements and select what best applies to you.

	Almost Always	Some of the time	Hardly Ever
I am satisfied that I can turn to my family for help when something is troubling me	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
I am satisfied with the way my family talks over things with me and shares problems with me	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
I am satisfied that my family accepts and supports my wishes to take on new activities or directions	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
I am satisfied with the way my family expresses affection and responds to my emotions, such as anger, sorrow and love	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
I am satisfied with the way my family and I share time together	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>



### Your Child's Strengths and Difficulties

*We would now like to ask about the strengths and difficulties of your **child with intellectual disability**. If there are any items that do not apply to your child then please select 'Not True'.*

57.

## Strengths and Difficulties Questionnaire

For each item, please mark the box for Not True, Somewhat True or Certainly True. It would help us if you answered all items as best you can even if you are not absolutely certain or the item seems daft! Please give your answers on the basis of the child's behaviour over the last six months.

	Not true	Somewhat true	Certainly true
Considerate of other people's feelings	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
Restless, overactive, cannot stay still for long	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
Often complains of headaches, stomach-aches or sickness	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
Shares readily with other children (treats, toys, pencils etc.)	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
Often has temper tantrums or hot tempers	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
Rather solitary, tends to play alone	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
Generally obedient, usually does what adults request	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
Many worries, often seems worried	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
Helpful if someone is hurt, upset or feeling ill	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
Constantly fidgeting or squirming	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
Has at least one good friend	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
Often fights with other children or bullies them	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
Often unhappy, down-hearted or tearful	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
Generally liked by other children	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
Easily distracted, concentration wanders	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
Nervous or clingy in new situations, easily loses confidence	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
Kind to younger children	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
Often lies or cheats	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
Picked on or bullied by other children	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
Often volunteers to help others (parents, teachers, other children)	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
Thinks things out before acting	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
Steals things from home, school or elsewhere	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
Gets on better with adults than with other children	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
Many fears, easily scared	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
Sees tasks through to the end, good attention span	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>

## Relationship with your child with intellectual disability

*We would now like to ask you about your relationship with your child with intellectual disability.*

58. Please reflect on the degree to which each of the following statements currently applies to your relationship with your child with intellectual disability. Using the scale below, select one appropriate answer for each item.

	Definitely does not apply	Not really	Neutral, not sure	Applies somewhat	Definitely applies
I share an affectionate relationship with this child	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
This child and I always seem to be struggling with each other	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
If upset, this child will seek comfort from me	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
This child is uncomfortable with physical affection or touch from me	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
This child values his/her relationship with me	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
When I praise this child, he/she beams with pride	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
This child spontaneously shares information about himself/herself	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
This child easily becomes angry with me	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
It is easy to be in tune with what this child is feeling	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
This child remains angry or is resistant after being disciplined	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
Dealing with this child drains my energy	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
When this child is in a bad mood, I know we're in for a long difficult day	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
This child's feelings towards me can be unpredictable or change suddenly	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
This child is sneaky or manipulative with me	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
This child openly shares his/her feelings and experiences with me	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>

## Parenting your child with intellectual disability

*We would now like to ask you some questions about your approach to parenting your child with intellectual disability.*

59. The following are a number of statements about your approach to parenting your child with intellectual disability. Please rate each item as to how often it typically occurs in your home.

	Never	Almost Never	Sometimes	Often	Always
You let your child know when he/she is doing a good job with something	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
You threaten to punish your child and then do not actually punish him/her	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
You reward or give something extra to your child for obeying you or behaving well	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
Your child talks you out of being punished after he/she has done something wrong	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
You feel that getting your child to obey you is more trouble than it's worth	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
You compliment your child when he/she does something well	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
You praise your child if he/she behaves well	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
You let your child out of a punishment early (e.g., lift restrictions earlier than you originally said.)	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
You hug or kiss your child when he/she has done something well	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
The punishment you give your child depends on your mood	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
Your child is not punished when he/she has done something wrong	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
You tell your child that you like it when he/she helps around the house	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>

*We would now like to ask you about how often you do particular activities with your child with intellectual disability.*

60. The following are a number of statements about you and your child with intellectual disability. Please rate each item as to how often it has typically occurred during the past six months

	Not at all	Less often than once or twice a month	Once or twice a month	Several times a week	Everyday
How often do you read or share a story with your child?	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
How often do you and your child sing together?	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
How often do you and your child play a game together?	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
How often do you and your child go out together for enjoyment? (rather than as a chore e.g. appointments/ food shopping etc.)	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
How often do you and your child watch TV together?	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>

61. Does your child with intellectual disability have at least one sibling between the ages of 4 to 15?

☐ Yes

☐ No

## Sibling Strengths and Difficulties

***We would now like to ask about the strengths and difficulties of one of the siblings of your child with intellectual disability.***

***If there is more than one sibling between the ages of 4 to 15 please choose the sibling closest in age to your child with intellectual disability.***

62. Age of sibling in years and months

	Years	Months
Age	<input type="text"/>	<input type="text"/>

63. Sibling gender

- ☐ Male
- ☐ Female

64. Does this sibling have a longstanding illness, disability or infirmity? By longstanding we mean anything that has troubled them over a period of time or is likely to affect them over a period of time?

- ☐ No
- ☐ Yes

65. Does this sibling live in the same household as your child with intellectual disability?

- ☐ All of the time
- ☐ Some of the time
- ☐ None of the time

66. Please give details of the sibling's longstanding illness, disability or infirmity.



We would now like to ask about the strengths and difficulties of the **sibling of your child** with intellectual disability. If there are any items that do not apply to your child then please select 'Not True'.

67.

### Strengths and Difficulties Questionnaire

For each item, please mark the box for Not True, Somewhat True or Certainly True. It would help us if you answered all items as best you can even if you are not absolutely certain or the item seems daft! Please give your answers on the basis of the child's behaviour over the last six months.

	Not true	Somewhat true	Certainly true
Considerate of other people's feelings	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
Restless, overactive, cannot stay still for long	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
Often complains of headaches, stomach-aches or sickness	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
Shares readily with other children (treats, toys, pencils etc.)	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
Often has temper tantrums or hot tempers	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
Rather solitary, tends to play alone	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
Generally obedient, usually does what adults request	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
Many worries, often seems worried	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
Helpful if someone is hurt, upset or feeling ill	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
Constantly fidgeting or squirming	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
Has at least one good friend	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
Often fights with other children or bullies them	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
Often unhappy, down-hearted or tearful	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
Generally liked by other children	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
Easily distracted, concentration wanders	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
Nervous or clingy in new situations, easily loses confidence	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
Kind to younger children	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
Often lies or cheats	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
Picked on or bullied by other children	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
Often volunteers to help others (parents, teachers, other children)	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
Thinks things out before acting	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>

	Not true	Somewhat true	Certainly true
Steals things from home, school or elsewhere	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
Gets on better with adults than with other children	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
Many fears, easily scared	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
Sees tasks through to the end, good attention span	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>

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68. We would now like you to tell us about the relationship between your child with intellectual disability and their sibling you have just told us about.

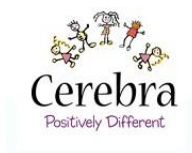
	Hardly at all	Not too much	Somewhat	Very much	Extremely much
How much do the sibling and the child tell each other everything?	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
How much do the sibling and the child share secrets and private feelings?	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
How much do the sibling and the child go places and do things together?	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
Some siblings play around and have fun with each other a lot, while other siblings play around and have fun with each other a little. How much do the sibling and the child play around and have fun with each other?	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
Some siblings care about each other a lot while other siblings don't care about each other that much. How much do the sibling and the child care about each other?	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
How much do the sibling and the child love each other?	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
How much do the sibling and the child disagree and quarrel with each other?	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
How much do the sibling and the child get mad and get in arguments with each other?	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
How much do the sibling and the child insult and call each other names?	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
How much are the sibling and the child mean to each other?	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>

Thank you for completing the 1000 Families survey. If you wish to forward this survey onto other parents, this would be greatly appreciated.

Please click "done" to finish

For updates on this study and other topics related to families of children with intellectual disability please visit our [Facebook](#) and [Twitter](#) page.

## Appendix F: Primary caregiver survey (Paper version)



# Cerebra 1,000 Families study

## Primary caregiver survey

*Please enter the following details below so that your survey responses can be matched with any other respondents from your household.*

### 1. Child with intellectual disability

First name of your child with intellectual disability:	
Surname of your child with intellectual disability:	

### 2. Your postcode

### 3. If your child lives at a different address to you, please enter your child's postcode

## Contacting you about the telephone interview

### 4. I agree to participate in a telephone interview with a researcher (as described in the information sheet).

Please select <u>ONE</u>	✓
Yes (Please complete Questions 5,6 and 7)	
No (Please go to Question 8)	

### 5. Please enter the following details so you can be contacted by a member of the research team to arrange a time for the telephone interview.

First name:	
Surname:	

**6. Telephone number**

Home telephone number:	
Mobile telephone number:	

**7. Email address**

--

Contacting you in 2 years' time

**8. I agree that the research team can contact me in 2 years' time to invite me to participate in the follow up study.**

Please select <u>ONE</u>	✓
Yes (Please complete Questions 9, 10 and 11)	
No (Please go to Question 12)	

**9. Please provide the following details so that we can contact you for the follow-up study in 2 years' time.**

First name:	
Surname:	
Address Line 1:	
Address Line 2:	
City/Town:	
Postcode:	

**10. Telephone number**

Home telephone number:	
------------------------	--

Mobile telephone number:	
--------------------------	--

11. Email

address

### Email updates

**12. I wish to be kept up to date by email with the progress and findings of the study and other research about families of individuals with intellectual disability.**

Please select <u>ONE</u>	✓
No	
Yes (Please provide your email address below)	

### Questions about you and your child with Intellectual Disability

**13. Is your child with intellectual disability male or female?**

Please select <u>ONE</u>	✓
Male	
Female	

### Questions about you

**14. Please indicate your relationship to the child with intellectual disability.**

Please select <u>ONE</u>	✓
Biological mother	
Biological father	
Adoptive mother	
Adoptive father	
Stepmother	
Stepfather	
Foster mother	
Foster father	
Grandmother	
Grandfather	

Other (please describe)

**15. How do you identify your gender?**

Please select <u>ONE</u>	✓
Male	
Female	
Trans	
Prefer not to answer	

**16. How would you describe your ethnicity?**

Please select <u>ONE</u>	✓
Asian/Asian British: Indian	
Asian/Asian British: Pakistani	
Asian/Asian British: Bangladeshi	
Asian/Asian British: Chinese	
Asian other (please describe below)	

Black/African/Black British: African	
Black/African/Black British: Caribbean	
Black other (please describe below)	

Mixed/multiple ethnic groups: White and Black Caribbean	
Mixed/multiple ethnic groups: White and Black African	
Mixed/multiple ethnic groups: White and Asian	
Mixed other (please describe below)	

Other Ethnic group: Arab	
Ethnic other (please describe)	

White: English/Welsh/Scottish/Northern Irish/British	
White: Irish	
White: Travelling community	
White: Other (Please describe below)	

Any other ethnic background (Please describe below)	



**17. Please select the highest level of your educational qualifications**

Please select <u>ONE</u>	✓
No qualifications	
Some GCSEs passes or equivalent	
5 or more GCSEs at A*-C or equivalent	
5 A/AS Levels or equivalent	
Higher Education but below degree level	
Degree (e.g. BA, BSC, MA)	
Don't know	

**18. Please select one option which best describes your status**

Please select <u>ONE</u>	✓
In a job and currently working for an employer	
On maternity/paternity /parental leave from a job	
Self-employed	
A Full time student	
Doing voluntary work	
Looking after home and family	
Unemployed	
Do something else (please specify below)	

**19. How is your health in general?**

Please select <u>ONE</u>	✓
Very good	
Good	
Fair	
Bad	
Very bad	

**20. Do you have a longstanding illness, disability or infirmity? By longstanding we mean anything that has troubled you over a period of time or is likely to affect you over a period of time?**

Please select <u>ONE</u>	✓
No	
Yes	

**Questions about your household**

**21. In total how many people currently live in your home (including yourself)?**

	<b>Adults</b>	<b>Children</b>
--	---------------	-----------------

Number		
--------	--	--

*Data from research with families with a family member with a disability has shown that a family's financial resources are important in understanding family member's views and experiences. With this in mind, we would be grateful if you could answer the additional question below. We are not interested in exactly what your family income is, but we would like to be able to look at whether people with different levels of financial resources have different experiences.*

**22. What is your total weekly household income (after any deductions e.g. income tax), including income from paid work, pension, Social Services Benefits (e.g. Job Seekers Allowance, DLA, Carers' Allowance, Attendance Allowance, Tax Credits, Housing Benefits, Pension Credits) etc.?**

Please select <u>ONE</u>	✓
£200 or less	
Between £200 and £300	
Between £300 and £400	
Between £400 and £500	
Between £500 and £600	
Between £600 and £700	
Between £700 and £800	
Between £800 and £900	
Over £1000	

**23. How well would you say you [and your husband/wife/partner] are managing financially these days?**

Would you say you are ...

Please select <u>ONE</u>	✓
living comfortably?	
doing alright?	
just about getting by?	
finding it quite difficult?	
finding it very difficult?	

**24. Suppose you only had one week to raise £2000 for an emergency, which of the following best describes how hard it would be for you to get that money?**

Please select <u>ONE</u>	✓
I could easily raise the money	

I could raise the money, but it would involve some sacrifices (e.g. reduced spending, selling a possession)	
I would have to do something drastic to raise the money (e.g. selling an important possession)	
I don't think I could raise the money	

### Questions about your child with intellectual disability

*We would now like to ask you some questions about your child with intellectual disability who you named earlier in the survey.*

#### **25. The date of birth of your child with intellectual disability**

DD  MM  YY

#### **26. Please select which of the conditions below professionals have diagnosed in relation to your child with intellectual disability (select ALL that apply)**

Select <u>ALL</u> that apply	✓
Learning disability/learning difficulty	
Autism/Autistic Spectrum Disorder /Autistic Spectrum Condition/Asperger's Syndrome	
Down syndrome	
Global Developmental delay	
Cerebral palsy	
Other genetic syndrome/diagnosis (please describe below)	

#### **27. Please state if your child with intellectual disability has:**

Please select <u>ONE</u>	✓
<b>A mild/moderate intellectual disability</b> Children with a mild to moderate intellectual disability can typically communicate and look after themselves well, but may take a bit longer to learn new skills compared to other children of the same age.	
<b>A Severe/profound intellectual disability</b> Children with a severe to profound intellectual disability are likely to have complex and multiple difficulties which require extensive support to learn and carry out daily activities.	

#### **28. Does your child with intellectual disability have a visual impairment?**

Please select <u>ONE</u>	✓
Yes	
No	

**29. Does your child with intellectual disability have a hearing impairment?**

Please select <u>ONE</u>	✓
Yes	
No	

**30. Does your child with intellectual disability currently have epileptic seizures?**

Please select <u>ONE</u>	✓
Yes (Please go to Question 32)	
No (Please go to Question 31)	

**31. Has your child with intellectual disability ever had an epileptic seizure in the past?**

Please select <u>ONE</u>	✓
Yes	
No	

**32. Does your child with intellectual disability have any mobility problems?**

Please select <u>ONE</u>	✓
Yes	
No	

**33. Does your child with intellectual disability have any other physical health problems?**

Please select <u>ONE</u>	✓
Yes	
No	

**34. Does your child with intellectual disability normally**

Please select <u>ONE</u>	✓
Live with you full-time? (Please go to Question 36)	
Live with you part-time? (Please got to Question 35)	

**35. Please state the approximate number of hours that your child lives with you on a weekly basis**

--

**36. What type of school does your child with intellectual disability usually attend?**

Please select <u>ONE</u>	✓
Mainstream school	
Mainstream school in either a special unit or resourced Special Educational Needs (SEN) provision	
Special school	
Home schooled	
Not currently in school	

**Questions about your experiences**

*The following statements ask about your experiences of having a child with intellectual disability.*

**37. Please respond to all questions by selecting the response which best describes how you feel about each statement.**

Please select <u>ONE</u> answer per statement	Strongly agree	Agree	Neither agree or disagree	Disagree	Strongly disagree
Since having this child I have grown as a person					
Having this child has helped me learn new things/skills					
Raising this child helps put my life into perspective					
Since having this child, my family has become closer to one another					
Since having this child, my family has become more tolerant and accepting					
Since having this child I have become more determined to face up to challenges					
Since having this child I have a greater understanding of other people					

*We would now like to ask you about the time you have to do leisure or social activities.*

**38. Has the ability to spend time doing leisure or social activities been affected by the assistance you give to your child with intellectual disability in any of the**

ways described below? (For all responses, this should be as a result of the caring and not for other reasons). Please select **ANY** that apply.

Please select <b>ANY</b> that apply	✓
Unable to socialise or take part in social or leisure activities at all (due to caring responsibilities)	
Reduced time with spouse or partner	
Reduced time with other family members	
Reduced time with friends	
Difficulties making new friends	
Reduced time spent doing sport or physical activity	
Reduced time spent doing pastime or hobby	

*The following questions ask about how you have been feeling during the past 30 days.*

**39. For each question, please click the number that best describes how often you had this feeling. During the past 30 days, about how often do you feel**

Please select <b>ONE</b> answer per statement	All of the time	Most of the time	Some of the time	A little of the time	None of the time
...nervous?					
...hopeless?					
...restless or fidgety?					
...so depressed that nothing could cheer you up?					
...that everything was an effort?					
...worthless?					

*We are now going to ask you about your satisfaction with life.*

**40. Here is a scale from 1-10 where '1' means that you are completely dissatisfied and '10' means that you are completely satisfied. All things considered, please could circle the number which corresponds with how satisfied or dissatisfied you are about the way your life has turned out so far.**

<b>1</b>	<b>2</b>	<b>3</b>	<b>4</b>	<b>5</b>	<b>6</b>	<b>7</b>	<b>8</b>	<b>9</b>	<b>10</b>
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**41. What is your current marital status?**

Please select <b>ONE</b>	✓
Married and living with spouse/civil partner (Please answer questions 42 and 43)	
Living with partner (Please answer questions 42 and 43)	
Divorced/Separated/Single/Widowed/Not currently living with partner (Please go to question 44)	

*We would now like to ask you some questions about your relationship with a spouse/partner.*

**42. How often do you and your [husband/wife/partner] disagree over issues relating to your child with intellectual disability?**

Please select <u>ONE</u>	✓
Never	
Less than once a week	
Once a week	
Several times a week	
Once a day	
More than once a day	
Can't say	

**43. Here is a scale from 1-7 where '1' means that you are very unhappy and '7' means that you are very happy. Please circle the number which best describes how happy or unhappy you are with your relationship with your spouse/partner, all things considered?**

<b>1</b> Very unhappy	<b>2</b>	<b>3</b>	<b>4</b>	<b>5</b>	<b>6</b>	<b>7</b> Very happy	<b>8</b> Can't say
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### Questions about your family

*We would now like to ask you about how satisfied you are with family life.*

**44. Please read the following statements and select what best applies to you.**

Please select <u>ONE</u> answer per statement	Almost always	Some of the time	Hardly ever
I am satisfied that I can turn to my family for help when something is troubling me			
I am satisfied with the way my family talks over things with me and shares problems with me			
I am satisfied that my family accepts and supports my wishes to take on new activities or directions			
I am satisfied with the way my family expresses affection and responds to my emotions, such as anger, sorrow and love			
I am satisfied with the way my family and I share time together			

## Your child's strengths and difficulties

We would now like to ask about the strengths and difficulties of your **child with intellectual disability**. If there are any items that do not apply to your child then please tick 'Not True'.

### 45. Strengths and Difficulties Questionnaire

For each item, please mark the box for Not True, Somewhat True or Certainly True. It would help us if you answered all items as best you can even if you are not absolutely certain or the item seems daft! Please give your answers on the basis of the child's behaviour over the **last six months**.

	Not true	Somewhat true	Certainly true
Considerate of other people's feelings			
Restless, overactive, cannot stay still for long			
Often complains of headaches, stomach-aches or sickness			
Shares readily with other children (treats, toys, pencils etc.)			
Often has temper tantrums or hot tempers			
Rather solitary, tends to play alone			
Generally obedient, usually does what adults request			
Many worries, often seems worried			
Helpful if someone is hurt, upset or feeling ill			
Constantly fidgeting or squirming			
Has at least one good friend			
Often fights with other children or bullies them			
Often unhappy, down-hearted or tearful			
Generally liked by other children			
Easily distracted, concentration wanders			
Nervous or clingy in new situations, easily loses confidence			
Kind to younger children			
Often lies or cheats			
Picked on or bullied by other children			
Often volunteers to help others (parents, teachers, other children)			
Thinks things out before acting			



Steals things from home, school or elsewhere			
Gets on better with adults than with other children			
Many fears, easily scared			
Sees tasks through to the end, good attention span			

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### Relationship with your child with intellectual disability

*We would now like to ask you about your relationship with your child with intellectual disability.*

**46. Please reflect on the degree to which each of the following statements currently applies to your relationship with your child with intellectual disability. Using the scale below, tick one appropriate answer for each item.**

Please select <u>ONE</u> answer per statement	Definitely does not apply	Not really	Neutral, not sure	Applies somewhat	Definitely applies
I share an affectionate relationship with this child					
This child and I always seem to be struggling with each other					
If upset, this child will seek comfort from me					
This child is uncomfortable with physical affection or touch from me					
This child values his/her relationship with me					
When I praise this child, he/she beams with pride					
This child spontaneously shares information about himself/herself					
This child easily becomes angry with me					

It is easy to be in tune with what this child is feeling					
This child remains angry or is resistant after being disciplined					
Dealing with this child drains my energy					
When this child is in a bad mood, I know we're in for a long difficult day					
This child's feelings towards me can be unpredictable or change suddenly					
This child is sneaky or manipulative with me					
This child openly shares his/her feelings and experiences with me					

### Parenting your child with intellectual disability

*We would now like to ask you some questions about your approach to parenting your child with intellectual disability.*

**47. The following are a number of statements about your approach to parenting your child with intellectual disability. Please rate each item as to how often it typically occurs in your home.**

Please select <u>ONE</u> answer per statement	Never	Almost Never	Sometimes	Often	Always
You let your child know when he/she is doing a good job with something					
You threaten to punish your child and then do not actually punish him/her					
You reward or give something extra to your child for obeying you or behaving well					
Your child talks you out of being punished after he/she has done something wrong					

You feel that getting your child to obey you is more trouble than it's worth					
You compliment your child when he/she does something well					
You praise your child if he/she behaves well					
You let your child out of a punishment early (e.g., lift restrictions earlier than you originally said.)					
You hug or kiss your child when he/she has done something well					
The punishment you give your child depends on your mood					
Your child is not punished when he/she has done something wrong					
You tell your child that you like it when he/she helps around the house					

*We would now like to ask you about how often you do particular activities with your child with intellectual disability.*

**48. The following are a number of statements about you and your child with intellectual disability. Please rate each item as to how often it has typically occurred during the past six months.**

Please select <u>ONE</u> answer per statement	Not at all	Less often than once or twice a month	Once or twice a month	Several times a week	Everyday
How often do you read or share a story with your child?					
How often do you and your child sing together?					
How often do you and your child play a game together?					

How often do you and your child go out together for enjoyment? (rather than as a chore e.g. appointments/ food shopping etc.)					
How often do you and your child watch TV together?					

**49. Does your child with intellectual disability have at least one sibling between the ages of 4 to 15?**

Please select <u>ONE</u>	✓
Yes (Please go to Question 50)	
No (Please go to the end of the survey)	

**Sibling strengths and difficulties**

*We would now like to ask about the strengths and difficulties of one of the siblings of your child with intellectual disability. If there is more than one sibling between the ages of 4 to 15 please choose the sibling closest in age to your child with intellectual disability.*

**50. Age of sibling in years and months**

Years  Months

**51. Sibling gender**

Please select <u>ONE</u>	✓
Male	
Female	

**52. Does this sibling have a longstanding illness, disability or infirmity? By longstanding we mean anything that has troubled them over a period of time or is likely to affect them over a period of time?**

Please select <u>ONE</u>	✓
No	
Yes (Please also answer question 54)	

**53. Does this sibling live in the same household as your child with intellectual disability?**

Please select <u>ONE</u>	✓
All of the time	
Some of the time	
None of the time	

**54. Please give details of the sibling's longstanding illness, disability or infirmity.**

--

*We would now like to ask about the strengths and difficulties of the **sibling of your child** with intellectual disability. If there are any items that do not apply to your child then please select 'Not True'.*

**55. Strengths and Difficulties Questionnaire**

For each item, please mark the box for Not True, Somewhat True or Certainly True. It would help us if you answered all items as best you can even if you are not absolutely certain or the item seems daft! Please give your answers on the basis of the child's behaviour over the **last six months**.

	<b>Not true</b>	<b>Somewhat true</b>	<b>Certainly true</b>
Considerate of other people's feelings			
Restless, overactive, cannot stay still for long			
Often complains of headaches, stomach-aches or sickness			
Shares readily with other children (treats, toys, pencils etc.)			
Often has temper tantrums or hot tempers			
Rather solitary, tends to play alone			
Generally obedient, usually does what adults request			
Many worries, often seems worried			
Helpful if someone is hurt, upset or feeling ill			
Constantly fidgeting or squirming			
Has at least one good friend			
Often fights with other children or bullies them			
Often unhappy, down-hearted or tearful			

Generally liked by other children			
Easily distracted, concentration wanders			
Nervous or clingy in new situations, easily loses confidence			
Kind to younger children			
Often lies or cheats			
Picked on or bullied by other children			
Often volunteers to help others (parents, teachers, other children)			
Thinks things out before acting			
Steals things from home, school or elsewhere			
Gets on better with adults than with other children			
Many fears, easily scared			
Sees tasks through to the end, good attention span			

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**56. We would now like you to tell us about the relationship between your child with intellectual disability and their sibling you have just told us about.**

Please select <u>ONE</u> answer per statement	Hardly at all	Not too much	Somewhat	Very much	Extremely much
How much do the sibling and the child tell each other everything?					
How much do the sibling and the child share secrets and private feelings?					
How much do the sibling and the child go places and do things together?					
Some siblings play around and have fun with each other a lot, while other siblings play around and have fun with each other a little. How much do the sibling and the child play around and have fun with each other?					
Some siblings care about each other a lot while other siblings don't care about each other that much. How much do the sibling and the child care about each other?					
How much do the sibling and the child love each other?					

How much do the sibling and the child disagree and quarrel with each other?					
How much do the sibling and the child get mad and get in arguments with each other?					
How much do the sibling and the child insult and call each other names?					
How much do the sibling and the child mean to each other?					

### **End of survey**

Thank you for completing the 1,000 Families survey. **Please return the survey with your consent form using the pre-post envelope in your pack.**

If you wish to forward this survey onto other parents, this would be greatly appreciated. The link to the online survey is [www.surveymonkey.com/r/1000families](http://www.surveymonkey.com/r/1000families) or alternatively a paper copy can be requested by emailing: [familyresearch@warwick.ac.uk](mailto:familyresearch@warwick.ac.uk).

For updates on this study and other topics related to families of children with intellectual disability please visit our Facebook and Twitter pages.



[www.facebook.com/FamilyRG1](http://www.facebook.com/FamilyRG1)



[@Family\\_RG1](https://twitter.com/Family_RG1)

## Appendix G: Ethical amendment confirmation

**From:** [nrescommittee.westmidlands-southbirmingham@nhs.net](mailto:nrescommittee.westmidlands-southbirmingham@nhs.net)  
 [mailto:nrescommittee.westmidlands-southbirmingham@nhs.net]  
**Sent:** 15 February 2017 10:43  
**To:** [M.Jess@warwick.ac.uk](mailto:M.Jess@warwick.ac.uk)  
**Cc:** [r.hastings@warwick.ac.uk](mailto:r.hastings@warwick.ac.uk); [wmssponsorship@warwick.ac.uk](mailto:wmssponsorship@warwick.ac.uk)  
**Subject:** IRAS 169882. Confirmation of REC Validation and Categorisation of Amendment

Dear Mikeda Jess,

<b>IRAS Project ID:</b>	<b>169882</b>
<b>REC Reference:</b>	<b>15/WM/0267</b>
<b>Short Study Title:</b>	<b>The 1,000 Families Study</b>
<b>Date complete amendment submission received:</b>	<b>13 February 2017</b>
<b>Amendment No./ Sponsor Ref:</b>	<b>SA 1</b>
<b>Amendment Date:</b>	<b>13 February 2017</b>
<b>Amendment Type:</b>	<b>Substantial</b>

Thank you for submitting the above referenced amendment. I am pleased to confirm that this amendment has been submitted to the REC for ethical review. Please find attached a copy of the validation letter.

### Categorisation of Amendment

In line with the [UK Process for Handling UK Study Amendments](#) I can confirm that this amendment has been categorised as:

- **Category A** - An amendment that has implications for, or affects, ALL participating NHS organisations

You should now provide this email, together with the amended documentation, to the research management support offices **and** local research teams at your participating NHS organisations in England.

If you have participating NHS organisations in Northern Ireland, Scotland and/or Wales, you should communicate directly with the relevant research teams to prepare them for implementing the amendment, as per the instructions below. You do not need to provide this email or your amended documentation to their research management



support offices, as we will pass these to the relevant national coordinating functions who will do this on your behalf.

Subject to the three conditions below, you will be able to implement the amendment at your participating NHS organisations in England **35 days after you notify them of the amendment**. A template email to notify participating NHS organisations in England is provided [here](#).

- You may not implement this amendment until and unless you receive all required regulatory approvals, including REC favourable opinion, (for participating organisations in England, this includes receiving confirmation of HRA Approval for the amendment). You should provide regulatory approvals to the research management support offices and local research teams at your participating NHS organisations in England, plus to local research teams at any participating NHS organisations in Northern Ireland, Scotland or Wales\*.
- You may not implement this amendment at any participating NHS organisations which inform you within the 35 day period that they require additional time to consider the amendment, until they notify you that the considerations have been satisfactorily completed.
- You may not implement this amendment at any participating NHS organisation that informs you that it is no longer able to undertake this study.

**Note:** you may only implement changes described in the amendment notice or letter.

If you receive required regulatory approvals (for participating organisations in England, this includes confirmation that the amendment has been granted HRA Approval) after the 35 days have passed, you may then immediately implement this amendment at all participating NHS organisations that have not requested additional review time, or are no longer able to undertake this study.

There is no need for you to receive a letter of confirmation from the participating organisation that the amendment can be implemented, as the intended date of implementation is communicated through the above process. However, you may be able to implement this amendment ahead of the 35 day deadline, if all necessary regulatory approvals are in place and the participating organisation has confirmed that the amendment may be implemented ahead of the 35 day date.

\* Where the study involves NHS organisations in Northern Ireland, Scotland or Wales, the HRA will forward regulatory approvals to the relevant national coordinating function to distribute to their research management support offices.

Please do not hesitate to contact me if you require further information.

Kind regards

Daniella Sarno

REC Assistant

Health Research Authority

HRA, Ground Floor, Skipton House, 80 London Road, London, SE1 6LH

E: [hra.amendments@nhs.net](mailto:hra.amendments@nhs.net)

[www.hra.nhs.uk](http://www.hra.nhs.uk)

The HRA is keen to know your views on the service you received – our short feedback form is available [here](#)

## Appendix H: Ethics amendment protocol

### IRAS Project Filter

The integrated dataset required for your project will be created from the answers you give to the following questions. The system will generate only those questions and sections which (a) apply to your study type and (b) are required by the bodies reviewing your study. Please ensure you answer all the questions before proceeding with your applications.

Please complete the questions in order. If you change the response to a question, please select 'Save' and review all the questions as your change may have affected subsequent questions.

**Please enter a short title for this project** (maximum 70 characters)

The 1,000 Families Study

**1. Is your project research?**

☒ Yes ☐ No

**2. Select one category from the list below:**

- ☐ Clinical trial of an investigational medicinal product
- ☐ Clinical investigation or other study of a medical device
- ☐ Combined trial of an investigational medicinal product and an investigational medical device
- ☐ Other clinical trial to study a novel intervention or randomised clinical trial to compare interventions in clinical practice
- ☐ Basic science study involving procedures with human participants
- ☒ Study administering questionnaires/interviews for quantitative analysis, or using mixed quantitative/qualitative methodology
- ☐ Study involving qualitative methods only
- ☐ Study limited to working with human tissue samples (or other human biological samples) and data (specific project only)
- ☐ Study limited to working with data (specific project only)
- ☐ Research tissue bank
- ☐ Research database

**If your work does not fit any of these categories, select the option below:**

☐ Other study

**2a. Please answer the following question(s):**

- a) Does the study involve the use of any ionising radiation? ☐ Yes ☒ No
- b) Will you be taking new human tissue samples (or other human biological samples)? ☐ Yes ☒ No
- c) Will you be using existing human tissue samples (or other human biological samples)? ☐ Yes ☒ No

**3. In which countries of the UK will the research sites be located?** *(Tick all that apply)*

- ☒ England
- ☒ Scotland

- ☒ Wales  
☒ Northern Ireland

3a. In which country of the UK will the lead NHS R&D office be located:

- ☐ England  
☐ Scotland  
☐ Wales  
☐ Northern Ireland  
☒ This study does not involve the NHS

4. Which applications do you require?

- ☐ NHS/HSC Research and Development offices  
☐ Social Care Research Ethics Committee  
☒ Research Ethics Committee  
☐ Confidentiality Advisory Group (CAG)  
☐ National Offender Management Service (NOMS) (Prisons & Probation)

*For NHS/HSC R&D Offices in Northern Ireland, Scotland and Wales the CI must create NHS/HSC Site Specific Information forms, for each site, in addition to the study wide forms, and transfer them to the PIs or local collaborators.*

*For participating NHS organisations in England different arrangements apply for the provision of site specific information. Refer to IRAS Help for more information.*

5. Will any research sites in this study be NHS organisations?

- ☐ Yes ☒ No

6. Do you plan to include any participants who are children?

- ☐ Yes ☒ No

7. Do you plan at any stage of the project to undertake intrusive research involving adults lacking capacity to consent for themselves?

- ☐ Yes ☒ No

*Answer Yes if you plan to recruit living participants aged 16 or over who lack capacity, or to retain them in the study following loss of capacity. Intrusive research means any research with the living requiring consent in law. This includes use of identifiable tissue samples or personal information, except where application is being made to the Confidentiality Advisory Group to set aside the common law duty of confidentiality in England and Wales. Please consult the guidance notes for further information on the legal frameworks for research involving adults lacking capacity in the UK.*

8. Do you plan to include any participants who are prisoners or young offenders in the custody of HM Prison Service or who are offenders supervised by the probation service in England or Wales?

- ☐ Yes ☒ No

9. Is the study or any part of it being undertaken as an educational project?

- ☒ Yes ☐ No

Please describe briefly the involvement of the student(s):

Three PhD students will work on data collection for this survey. Each will be examining and reporting data from a different area of the survey, with their own research questions, to contribute towards their PhD theses.

**9a. Is the project being undertaken in part fulfilment of a PhD or other doctorate?**

☒ Yes ☐ No

**10. Will this research be financially supported by the United States Department of Health and Human Services or any of its divisions, agencies or programs?**

☐ Yes ☒ No

**11. Will identifiable patient data be accessed outside the care team without prior consent at any stage of the project (including identification of potential participants)?**

☐ Yes ☒ No

**NOTICE OF SUBSTANTIAL AMENDMENT**

*Please use this form to notify the main REC of substantial amendments to all research other than clinical trials of investigational medicinal products (CTIMPs).  
The form should be completed by the Chief Investigator using language comprehensible to a lay person.*

**Details of Chief Investigator:**

	Title	Forename/Initials	Surname
	Professor	Richard	Hastings
Work Address	CEDAR (Centre for Educational Development Appraisal and Research)		
	University of Warwick		
	Coventry UK		
PostCode	CV47AL		
Email	r.hastings@warwick.ac.uk		
Telephone	+44 (0)2476 522197		
Fax			

**For guidance on this section of the form refer to the guidance**

<b>Full title of study:</b>	The 1,000 families study: Well-being in families of children with intellectual disability
<b>Lead sponsor:</b>	University of Warwick
<b>Name of REC:</b>	West Midlands - South Birmingham
<b>REC reference number:</b>	15/WM/0267

**Additional reference number(s):**

Ref.Number	Description	Reference Number
Sponsor reference number		REGO-2015-1604

**Name of lead R&D office:**

<b>Date study commenced:</b>	26th November 2015
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**Protocol reference (if applicable), current version and date:**

<b>Amendment number and date:</b>	Version 1 (13/01/2017)
-----------------------------------	------------------------

**Type of amendment**

(a) Amendment to information previously given in IRAS

☒ Yes ☐ No

If yes, please refer to relevant sections of IRAS in the "summary of changes" below.

A17-1 inclusion criteria upper age for children increased to 15 years 11 months

**(b) Amendment to the protocol**

☒ Yes ☐ No

If yes, please submit either the revised protocol with a new version number and date, highlighting changes in bold, or a document listing the changes and giving both the previous and revised text.

Revised protocol attached with updates to change upper age for inclusion. We have also taken the opportunity to update research team and other contact information. Amended protocol showing all tracked changes attached.

**(c) Amendment to the information sheet(s) and consent form(s) for participants, or to any other supporting documentation for the study**

☒ Yes ☐ No

If yes, please submit all revised documents with new version numbers and dates, highlighting new text in bold.

Upper age for inclusion amended, and updated information about an independent research contact for carers wishing to ask questions about the study.

**Is this a modified version of an amendment previously notified and not approved?**

☐ Yes ☒ No

**Summary of changes**

Briefly summarise the main changes proposed in this amendment. Explain the purpose of the changes and their significance for the study.

If this is a modified amendment, please explain how the modifications address the concerns raised previously by the ethics committee.

If the amendment significantly alters the research design or methodology, or could otherwise affect the scientific value of the study, supporting scientific information should be given (or enclosed separately). Indicate whether or not additional scientific critique has been obtained.

At present, we are recruiting caregivers raising children with Intellectual Disability (ID) who are between the ages of 4 years to 11 years and 11 months. We would like to change our inclusion criteria to extend the maximum age of the child with ID at recruitment 15 years and 11 months. There are two main reasons for this change. First, although recruitment has been successful (approximately 600 families to date), the recruitment rate has been slower than anticipated. To be able to complete recruitment within a reasonable time frame, we need to increase the recruitment rate. Extending the potential pool of participants would be one simple way to achieve this. Second, during recruitment, a large minority of potential participants have been parents of children between the ages of 12 and 16 years. They are currently not able to take part in the study, but have expressed an interest to do so.

This change to the upper age for recruitment is the only amendment we are proposing. The revised documents include the Protocol, the text used for a simple advertisement and the information sheets for parents. The information sheets also include revised cosmetic appearance that matches the look of the information provided within the online survey. We have also taken the opportunity to update the contact information for a person not on the research team who is available to talk to parents about their potential participation in the research. Additionally, we have updated the complaints contact information on the information sheets.

No other changes have been made to the protocol, except for updating of contact information. This proposed amendment does not change the rationale/purpose for the study or any other element of the study procedure.

**Any other relevant information**

Applicants may indicate any specific issues relating to the amendment, on which the opinion of a reviewing body is sought.

**List of enclosed documents**

<i>Document</i>	<i>Version</i>	<i>Date</i>
Secondary caregiver study information	1.2	13/01/2017
Primary caregiver study information	1.2	13/01/2017
Project advertisement text	1.2	13/01/2017
Cerebra 1000 Families Project Protocol v1.1 6.1.17	1.1	13/01/2017

#### **Declaration by Chief Investigator**

1. *I confirm that the information in this form is accurate to the best of my knowledge and I take full responsibility for it.*
2. *I consider that it would be reasonable for the proposed amendment to be implemented.*

This section was signed electronically by Professor Richard Hastings on 10/02/2017 00:02.

Job Title/Post: Professor  
 Organisation: University of Warwick  
 Email: R.Hastings@warwick.ac.uk

#### **Declaration by the sponsor's representative**

*I confirm the sponsor's support for this substantial amendment.*

This section was signed electronically by Mrs Jane Prewett on 08/02/2017 12:12.

Job Title/Post: Head of Research Governance  
 Organisation: University of Warwick  
 Email: jane.prewett@warwick.ac.uk



## Appendix I: Recruitment flyers (pre and post age amendment)




# 1,000 Families study

**Are you the mother, father, or caregiver of a child aged 4-11 years who has a learning disability?**

If so, please take part in our new 'Cerebra 1000 Families study.' Your child might have other conditions - such as Autism, or any one of a number of genetic syndromes. As long as your child also has a learning disability, we would be very keen to hear from you.

**The Cerebra Family Research Group** at the University of Warwick, in collaboration with the research charity Cerebra, wishes to explore the experiences of **family members who live with a child with a learning disability in the UK.**

We are really keen to learn about the experiences of **at least one person** in the family. Most research studies only ask one person in the household to report on their family, however if **more than one family member** participates (especially, a mother and a father) we will be able to explore whether family members have similar or different experiences.

We want to make this the largest study ever of families of children with a learning disability in the UK



[www.facebook.com/FamilyRG1](https://www.facebook.com/FamilyRG1)



[@Family\\_RG1](https://twitter.com/Family_RG1)

This research will **further our understanding** of what it is like to raise a child with a learning disability in the UK and will **help shape future support** for families and children.

**To take part please go to:**

**[www.surveymonkey.com/r/1000families](https://www.surveymonkey.com/r/1000families)**

If you have any questions or would like to know more about the study, please contact the research team by email: [familyresearch@warwick.ac.uk](mailto:familyresearch@warwick.ac.uk) or telephone: 02476 524 139



## 1,000 Families study

**Are you the mother, father, or caregiver of a child aged between 4 and 15 years, 11 months who has a learning disability?**

If so, please take part in our new 'Cerebra 1000 Families study.' Your child might have other conditions - such as Autism, or any one of a number of genetic syndromes. As long as your child also has a learning disability, we would be very keen to hear from you.

The **Cerebra Family Research Group** at the University of Warwick, in collaboration with the research charity Cerebra, wishes to explore the experiences of **family members who live with a child with a learning disability in the UK.**

We want to make this the **largest** study ever of families of children with a learning disability in the UK

We are really keen to learn about the experiences of **at least one person** in the family. Most research studies only ask one person in the household to report on their family, however **if more than one family member** participates (especially a mother and a father) we will be able to explore whether family members have similar or different experiences.



[www.facebook.com/FamilyRG1](https://www.facebook.com/FamilyRG1)



@Family\_RG1

This research will **further our understanding** of what it is like to raise a child with a learning disability in the UK and will **help shape future support** for families and children.

**To take part please go to:**

**[www.surveymonkey.com/r/1000families](https://www.surveymonkey.com/r/1000families)**

If you have any questions or would like to know more about the study, please contact the research team by email: [familyresearch@warwick.ac.uk](mailto:familyresearch@warwick.ac.uk) or telephone: 02476 524 139

## Appendix J: Cerebra mail out to member database

### Cerebra 1,000 Families Study

If you are the mother, father or caregiver of children with a learning disability, this is your opportunity to inform research and policy for coming decades!

We understand that families play a vital role in their children's lives. By listening to what families tell us we are able to help families see past barriers and discover a better life together.

Together with the Cerebra Family Research Group at the University of Warwick we are exploring the experiences of family members who live with a child with a learning disability aged **4 to 11 years**. You can also take part if your child has a learning disability and other conditions such as Down syndrome, autism, or other genetic syndromes.

The study involves taking a short survey and we are really keen to hear from *at least* one person in the family. We would like to hear from fathers as well as mothers.

We want to make this the largest study ever of families of children with learning disability in the UK. The 1,000 families study will further understanding of what it is like to raise a child with a learning disability and will help shape future information and support for families and children – so please get involved.

**To find out more about the study and to complete the online survey please visit the website. If you prefer you can request a paper copy of the survey.**

If you have any questions please email: [familyresearch@warwick.ac.uk](mailto:familyresearch@warwick.ac.uk)

Thank you for taking the time to consider taking part in this important family research project. We would be very grateful if you could forward this email on to anyone else you think might be interested and help us to reach 1,000 families!

We're the charity dedicated to helping families with children with brain conditions discover a better life together. It's an incredibly rewarding journey for everyone involved. Why not be part of it? You never know what we'll discover together.

Best wishes



Tracy Elliott (Cerebra Head of Research)

## Appendix K: Recruitment postcard

# Cerebra 1,000 families study

What is it like raising a child with an intellectual (learning) disability in the UK? We want to hear from you about your experiences.





To take part visit  
[www.surveymonkey.com/r/1000families](http://www.surveymonkey.com/r/1000families)



### Take part in our survey

Our study will raise awareness of what it is like to raise a child with an intellectual (learning) disability in the UK and will shape decades of research and support for families.

---

For more information or to request a paper copy of the survey contact us at [familyresearch@warwick.ac.uk](mailto:familyresearch@warwick.ac.uk)


@family\_RG1


facebook.com/familyRG1

### Who can take part?



You can be a mother, father or family member of a child who is aged between 4 and 11 years AND has an intellectual (learning) disability (your child may have other conditions such as autism, or any one of a number of genetic syndromes)

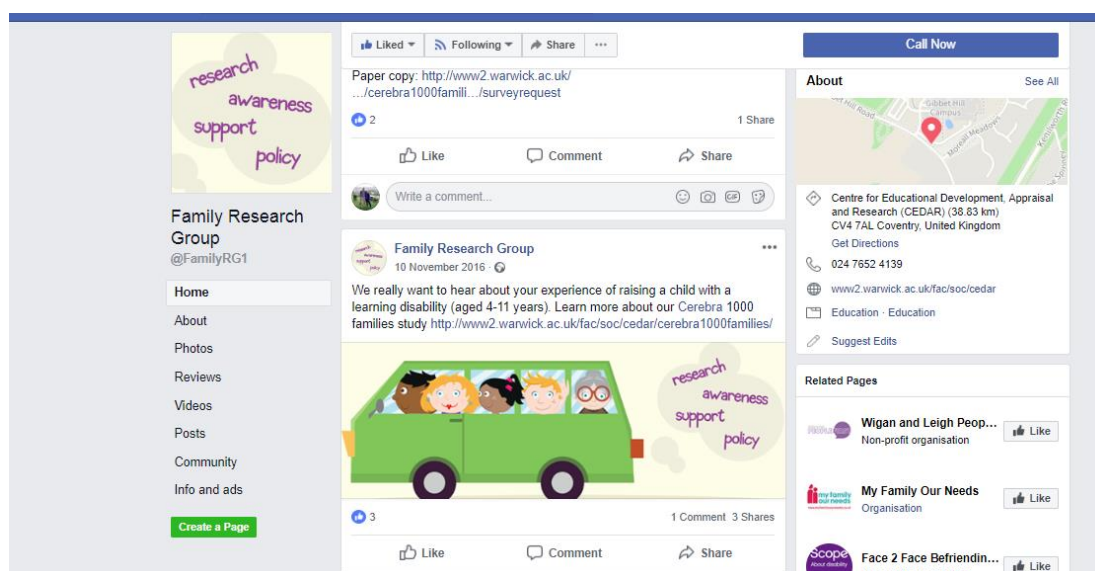
### What will it involve?



Online or paper survey (approx. 20 mins)

Telephone interview (optional and usually with the primary caregiver)

## Appendix L: Social media recruitment examples



**Appendix M: Dyadic Adjustment Scale (DAS-7)**  
**Hunsley, Pinsent, Lefebvre, Jamestaner, & Vito (1995)**

1-3 Most persons have disagreements in their relationships. Please indicate below the approximate extent of agreement or disagreement between you and your partner for each item on the following list.

5=Always Agree  
 4=Almost Always Agree  
 3=Occasionally Disagree  
 2=Frequently Disagree  
 1=Almost Always Disagree  
 0=Always Disagree

(Circle one number for each item)

1. Philosophy of life	5	4	3	2	1	0
2. Aims, goals, and things believed important	5	4	3	2	1	0
3. Amount of time spent together.	5	4	3	2	1	0

4-6 How often would you say the following events occur between you and your mate?

0=Never  
 1=Less than once a month  
 2=Once or twice a month  
 3=Once or twice a week  
 4=Once a day  
 5=More often (than once a day)

(Circle one number for each item)

4. Have a stimulating exchange of ideas	0	1	2	3	4	5
5. Calmly discuss something together	0	1	2	3	4	5
6. Work together on a project	0	1	2	3	4	5

7. The following represent different degrees of happiness in your relationship. The middle point, "happy," represents the degree of happiness of most relationships. Please circle the degree of happiness, all things considered, in your relationship.

Extremely Unhappy	Fairly Unhappy	A Little Unhappy	Happy	Very Happy	Extremely Happy	Perfect
0	1	2	3	4	5	6

## Appendix N: Hospital Anxiety and Depression Scale (HADS) Zigmond and Snaith (1983)

The following are questions about your health and wellbeing. Read each item and tick the reply which comes closest to how you have been feeling in the past week. Try not to take too long over your replies: your immediate reaction to each item will probably be more accurate than a long thought out response.

1. I feel tense or 'wound up'					
Most of the time	<input type="checkbox"/>	A lot of the time	<input type="checkbox"/>	From time to time, occasionally	<input type="checkbox"/> Not at all <input type="checkbox"/>
2. I still enjoy the things I used to enjoy					
Definitely as much	<input type="checkbox"/>	Not quite so much	<input type="checkbox"/>	Only a little	<input type="checkbox"/> Hardly at all <input type="checkbox"/>
3. I get a sort of frightened feeling as if something awful is about to happen					
Very definitely and quite badly	<input type="checkbox"/>	Yes, but not too badly	<input type="checkbox"/>	A little, but it doesn't worry me	<input type="checkbox"/> Not at all <input type="checkbox"/>
4. I can laugh and see the funny side of things					
As much as I always could	<input type="checkbox"/>	Not quite so much now	<input type="checkbox"/>	Definitely not so much now	<input type="checkbox"/> Not at all <input type="checkbox"/>
5. Worrying thoughts go through my mind					
A great deal of the time	<input type="checkbox"/>	A lot of the time	<input type="checkbox"/>	From time to time but not too often	<input type="checkbox"/> Only occasionally <input type="checkbox"/>
6. I feel cheerful					
Not at all	<input type="checkbox"/>	Not often	<input type="checkbox"/>	Sometimes	<input type="checkbox"/> Most of the time <input type="checkbox"/>
7. I can sit at ease and feel relaxed					
Definitely	<input type="checkbox"/>	Usually	<input type="checkbox"/>	Not often	<input type="checkbox"/> Not at all <input type="checkbox"/>
8. I feel as if I am slowed down					
Nearly all the time	<input type="checkbox"/>	Very often	<input type="checkbox"/>	Sometimes	<input type="checkbox"/> Not at all <input type="checkbox"/>
9. I get a sort of frightened feeling like 'butterflies' in the stomach					
Not at all	<input type="checkbox"/>	Occasionally	<input type="checkbox"/>	Quite often	<input type="checkbox"/> Very often <input type="checkbox"/>
10. I have lost interest in my appearance					
Definitely	<input type="checkbox"/>	I don't take so much care as I should	<input type="checkbox"/>	I may not take quite as much care	<input type="checkbox"/> I take just as much care as ever <input type="checkbox"/>
11. I feel restless as if I have to be on the move					
Very much indeed	<input type="checkbox"/>	Quite a lot	<input type="checkbox"/>	Not very much	<input type="checkbox"/> Not at all <input type="checkbox"/>
12. I look forward with enjoyment to things					
As much as ever I did	<input type="checkbox"/>	Rather less than I used to	<input type="checkbox"/>	Definitely less than I used to	<input type="checkbox"/> Hardly at all <input type="checkbox"/>
13. I get sudden feelings of panic					
Very often indeed	<input type="checkbox"/>	Quite often	<input type="checkbox"/>	Not very often	<input type="checkbox"/> Not at all <input type="checkbox"/>
14. I can enjoy a good book or radio or TV programme					
Often	<input type="checkbox"/>	Sometimes	<input type="checkbox"/>	Not often	<input type="checkbox"/> Very seldom <input type="checkbox"/>

**Appendix O: Family APGAR Scale**  
**Smilkstein (1978)**

	Almost Always	Some of the Time	Hardly Ever
1) I am satisfied that I can turn to my family for help when something is troubling me.			
2) I am satisfied with the way my family talks over things with me and shares problems with me.			
3) I am satisfied that my family accepts and supports my wishes to take on new activities or directions.			
4) I am satisfied with the way my family expresses affection and responds to my emotions, such as anger, sorrow, and love.			
5) I am satisfied with the way my family and I share time together.			

**Rating Scale:**

Almost Always = 2 pts.  
 Some of the Time = 1 pt.  
 Hardly Ever = 0 pts.

**Scoring:**

8-10 = Highly Functional  
 4-7 = Moderately Dysfunctional  
 0-3 = Dysfunctional

**Example:**

Total = 7 pts.  
 Moderately Dysfunctional



## Appendix P: Child-Parent Relationship Scale (CPRS) Pianta (1992)

Child: \_\_\_\_\_ Age: \_\_\_\_\_

Parent: \_\_\_\_\_

Please reflect on the degree to which each of the following statements currently applies to your relationship with your child. Using the scale below, circle the appropriate number for each item.

	Definitely does not apply 1	Not really 2	Neutral, not sure 3	Applies somewhat 4	Definitely applies 5
1. I share an affectionate, warm relationship with my child.	1	2	3	4	5
2. My child and I always seem to be struggling with each other.	1	2	3	4	5
3. If upset, my child will seek comfort from me.	1	2	3	4	5
4. My child is uncomfortable with physical affection or touch from me.	1	2	3	4	5
5. My child values his/her relationship with me.	1	2	3	4	5
6. My child appears hurt or embarrassed when I correct him/her.	1	2	3	4	5
7. My child does not want to accept help when he/she needs it.	1	2	3	4	5
8. When I praise my child, he/she beams with pride.	1	2	3	4	5
9. My child reacts strongly to separation from me.	1	2	3	4	5
10. My child spontaneously shares information about himself/herself.	1	2	3	4	5
11. My child is overly dependent on me.	1	2	3	4	5
12. My child easily becomes angry at me.	1	2	3	4	5
13. My child tries to please me.	1	2	3	4	5
14. My child feels that I treat him/her unfairly.	1	2	3	4	5
15. My child asks for my help when he/she really does not need help.	1	2	3	4	5
16. It is easy to be in tune with what my child is feeling.	1	2	3	4	5
17. My child sees me as a source of punishment and criticism.	1	2	3	4	5
18. My child expresses hurt or jealousy when I spend time with other children.	1	2	3	4	5
19. My child remains angry or is resistant after being disciplined.	1	2	3	4	5
20. When my child is misbehaving, he/she responds to my look or tone of voice.	1	2	3	4	5
21. Dealing with my child drains my energy.	1	2	3	4	5
22. I've noticed my child copying my behavior or ways of doing things.	1	2	3	4	5
23. When my child is in a bad mood, I know we're in for a long and difficult day.	1	2	3	4	5
24. My child's feelings toward me can be unpredictable or can change suddenly.	1	2	3	4	5
25. Despite my best efforts, I'm uncomfortable with how my child and I get along.	1	2	3	4	5
26. I often think about my child when at work.	1	2	3	4	5
27. My child whines or cries when he/she wants something from me.	1	2	3	4	5
28. My child is sneaky or manipulative with me.	1	2	3	4	5
29. My child openly shares his/her feelings and experiences with me.	1	2	3	4	5
30. My interactions with my child make me feel effective and confident as a parent.	1	2	3	4	5

**Appendix Q: Sibling Relationship Questionnaire – revised (SRQ brief parent-version)**

**Furman & Buhrmester (1985)**

Please select <u>ONE answer per statement</u>	Hardly at all	Not too much	Somewhat	Very much	Extremely much
How much do the sibling and the child tell each other everything?					
How much do the sibling and the child share secrets and private feelings?					
How much do the sibling and the child go places and do things together?					
Some siblings play around and have fun with each other a lot, while other siblings play around and have fun with each other a little. How much do the sibling and the child play around and have fun with each other?					
Some siblings care about each other a lot while other siblings don't care about each other that much. How much do the sibling and the child care about each other?					
How much do the sibling and the child love each other?					
How much do the sibling and the child disagree and quarrel with each other?					
How much do the sibling and the child get mad and get in arguments with each other?					
How much do the sibling and the child insult and call each other names?					
How much are the sibling and the child mean to each other?					

**Appendix R: Strengths and Difficulties Questionnaire (SDQ)**  
**Goodman (1997, 2005)**

For each item, please mark the box for Not True, Somewhat True or Certainly True. It would help us if you answered all items as best you can even if you are not absolutely certain or the item seems daft! Please give your answers on the basis of the child's behaviour over the **last six months**.

	<b>Not true</b>	<b>Somewhat true</b>	<b>Certainly true</b>
Considerate of other people's feelings			
Restless, overactive, cannot stay still for long			
Often complains of headaches, stomach-aches or sickness			
Shares readily with other children (treats, toys, pencils etc.)			
Often has temper tantrums or hot tempers			
Rather solitary, tends to play alone			
Generally obedient, usually does what adults request			
Many worries, often seems worried			
Helpful if someone is hurt, upset or feeling ill			
Constantly fidgeting or squirming			
Has at least one good friend			
Often fights with other children or bullies them			
Often unhappy, down-hearted or tearful			
Generally liked by other children			
Easily distracted, concentration wanders			
Nervous or clingy in new situations, easily loses confidence			
Kind to younger children			
Often lies or cheats			
Picked on or bullied by other children			
Often volunteers to help others (parents, teachers, other children)			
Thinks things out before acting			
Steals things from home, school or elsewhere			
Gets on better with adults than with other children			
Many fears, easily scared			
Sees tasks through to the end, good attention span			

**Appendix S: Kessler 6 (K6)**  
**Kessler et al. (2003)**

Date completed: \_\_\_\_ / \_\_\_\_ / \_\_\_\_

K6+

Provider: \_\_\_\_\_

Provider ID: \_\_\_\_\_

Please use gummed label if available Patient or Client Identifier: \_\_\_\_\_

Surname: _____	
Other names: _____	
Date of Birth: _____	Sex: _____
____ / ____ / ____	Male <input type="checkbox"/> Female <input type="checkbox"/>
Address: _____	

The following questions ask about how you have been feeling during the past 30 days. For each question, please circle the number that best describes how often you had this feeling.

Q1. During the past 30 days, about how often did you feel ...	All of the time	Most of the time	Some of the time	A little of the time	None of the time
a. ...nervous?	1	2	3	4	5
b. ...hopeless?	1	2	3	4	5
c. ...restless or fidgety?	1	2	3	4	5
d. ...so depressed that nothing could cheer you up?	1	2	3	4	5
e. ...that everything was an effort?	1	2	3	4	5
f. ...worthless?	1	2	3	4	5

**Appendix T: Impact of caregiving on carer**  
**Survey of Informal carers in households (2009/10)**

**Has your ability to spend time doing leisure or social activities been affected by the assistance you give to (NAMED PERSON CARED FOR) in any of the ways on this card? This includes seeing friends and family, spending time with a spouse or partner. (FOR ALL RESPONSES, THESE SHOULD BE AS A RESULT OF THE CARING AND NOT FOR OTHER REASONS). CODE ALL THAT APPLY**

1. Unable to socialise or take part in social or leisure activities at all (due to caring responsibilities)
2. Reduced time with spouse or partner
3. Reduced time with other family members
4. Reduced time with friends
5. Difficulties making new friends
6. Reduce time spent doing sport or physical activity
7. Reduced time spent doing pastime or hobby

## **Appendix U: Positive Gain Scale (PGS)**

### **Pit-ten Cate (2003)**

#### **POSITIVE GAIN SCALE**

- 1) Since having this child I feel I have grown as a person
- 2) Having this child has helped me to learn new things/skills
- 3) Raising this child helps putting life into perspective
- 4) Since having this child, my family has become closer to one another
- 5) Since having this child my family has become more tolerant and accepting
- 6) Since having this child I have become more determined to face up to challenges
- 7) Since having this child I have a greater understanding of other people.

All items are score on a 5-point Likert scale (1 = strongly agree, 5 = strongly disagree)

## **Appendix V: Additional statistical information (all analysis chapters)**

### Chapter 2 – Parental relationship satisfaction in families of children with Autism Spectrum Disorder (ASD): A multilevel analysis

Multilevel models (MLM) have the same assumptions as general linear models (GLM) such as linear relationships, homoscedasticity, and normal distribution of the residuals, however MLMs can account for non-independence of observations (Maas & Hox, 2004). The DV dyadic adjustment was normally distributed, as were IVs depression and child behaviour problems. Sibling behaviour problems had a positively skewed distribution, which was to be expected. A family socioeconomic position (SEP) composite was chosen over individual indicators of SEP because the aim of the study was to explore the relationship between relationship satisfaction and socioeconomic position at the family level (level 2), rather than the individual level (level 1).

Family data violates the assumption of independence of observations due to their hierarchical (nested) design (i.e. mothers and fathers in the same household are more likely to report in similar ways), however MLMs accommodate data which violate this assumption. Data in the study were structured in a 2-tier hierarchical data structure with individual variation at Level 1 and family variation at Level 2. We explored the intraclass correlation (ICC) which describes the proportion of variance that is common to each unit (Heck et al, 2014). The ICC demonstrated the need to conduct a multilevel analysis as the higher-level grouping (“family”) affected the estimates. A different variance-correlation matrix: Restricted Maximum Likelihood (REML) was specified to account for heterogeneity of variance.

### Chapter 3 – The psychological wellbeing of fathers with and without a child with Intellectual Disability: A population-based study

Descriptive statistics showed that the dependent variables, life satisfaction and work-family balance were normally distributed and so were analysed using general linear regression. General health, parenting competency, and parent-child closeness had negatively skewed distributions and so were dichotomised accordingly. General health scores were dichotomized into two groups: 0=poor health (scores of 2 or

lower) and 1=good health (scores of 3 or higher). Parenting competency scores were dichotomised into 0=low level of parenting competence (scores of 1 and 2) and 1=high level of parenting competence (scores of 3 or higher). Parent-child closeness was dichotomised into 0=not close (scores of 1 and 2) and 1=close (scores of 3 and 4). These were analysed using logistic regression.

Individual predictors of socioeconomic position (SEP) were favoured over a composite as we were particularly interested in the relationships between each predictor and the outcomes. Furthermore, recent research has indicated that a composite can mask these different associations (Totsika et al., 2016). The individual indicators (paternal education, employment status, and income poverty) were dichotomised into degree/no degree education, in employment /not in employment, and above or below the 60% national equivalised median income.

#### Chapter 4 – Family relationships and their associations with perceptions of family functioning in mothers of children with Intellectual Disability (ID)

The assumptions of SEM were explored prior to performing the analysis. The outcome variable family APGAR was normally distributed. Some outlying cases were excluded for some IVs, including relationship satisfaction, to improve normality. The sample was of a good size ( $N=431$ ). Models could therefore be estimated using Maximum Likelihood (ML) estimation. We did, however, have some missing data so Full Information Maximum Likelihood (FIML) was used to account for missing values to produce estimates which allowed for the fit of a model to an entire sample.

As the chi-square goodness of fit index is sensitive to large sample size (Hu & Bentler, 1999), we evaluated the fit of the model by consulting a number of goodness of fit indexes including: the Tucker Lewis Index (TLI), Comparative Fit Index (CFI), the Root Mean Square Error of Approximation (RMSEA), and Standardized Root Mean Square Residual (SRMR).

#### Chapter 5 – Psychological wellbeing in single mothers of children with Intellectual Disability (ID)



The two dependent variables (psychological distress and life satisfaction) were normally distributed and so general linear models were conducted. Predictors such as the positive gain scale (PGS), strengths and difficulties questionnaire (SDQ), impact on personal life, and the additional needs index, all were normally distributed and so were used as continuous variables in the models.

Measures which comprised the socioeconomic position (SEP) composite (maternal education, maternal employment, subjective poverty, and hardship) were dichotomised into two groups (0/1). Mother education and employment predictors were dichotomised into degree/no degree education, and in employment /not in employment. The subjectivity poverty measure was dichotomised into 0=financially struggling (scores 4 and 5) and 1=financially managing (scores 1 to 3) based on its distribution and because this dichotomy has been used in previous research (Totsika et al., 2014). The hardship variable was dichotomised into 0=could not raise the money (scores of 4) and 1=could raise the money (scores of 1 to 3) as frequencies showed that over 50% of mothers selected the lowest category 'I could not raise the money.'

To decide whether the four indicators should be used together in a SEP or entered into the model separately, correlations with the outcomes were explored, checking whether they were all in the same direction and had small effect sizes. An OECD income poverty measure, which has been a reliable and informative indicator in previous research in the field, was also not readily available at the time of the analysis.

A correlation matrix of predictor variables was conducted to look for evidence of multicollinearity.